Pediatric Evaluation of Disability Inventory

-The Dutch Adaptation-

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'Quality of life is in the eye of the beholder'
uit: R. C. Ziller (1974) Self orientation and quality of life
Social Indicators Research, vol. 1:301-327

Aan mijn moeder
Voor Daan, Lot en Carry

Contents

Introduction

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Chapter 1

Disablement Process

Over the last few decades there have been two leading constructs in disability research. The first one is the 'Disablement Concept' of Nagi[1,2] the second one is the 'International Classification of Impairments, Disabilities, and Handicaps (ICIDH)[3]. Both were developed to clarify the consequences of diseases, disorders, and injuries on mankind. Although the ICIDH, recently evolved in ICIDH-2[4], is more widely known, Nagi's concept will be used in this dissertation for two reasons. First, the theoretical construct of the 'Pediatric Evaluation of Disability Inventory' (PEDI)[5,6], the subject of this dissertation, is mainly derived from Nagi's disablement concept, and second, it proves to be a useful concept in physical therapy research[7-10].

In the mid-sixties, the sociologist Saad Nagi described the 'Disablement Concept' in which he distinguishes four levels: active pathology, impairment, functional limitation, and disability. This concept involves physical as well as cognitive functions. Active pathology refers to 'a state of mobilisation of the body's defenses and coping mechanisms, resulting from infection, metabolic imbalances, traumatic injury, or other etiology'[1,2]. Nagi used the term impairments for 'any loss or abnormalities at the tissue, organ or body system level[1,2]. Functional limitations expresses the 'limitations in performance at the level of the whole person'[1,2]. The difference between impairments and functional limitations is the level at which these limitations are manifest themselves. A contracture in the hip joint can be viewed as an impairment, and may or may not limit walking outdoors. A limitation of walking is considered a functional limitation. Disability refers to an 'inability in performing socially defined roles and tasks within a sociocultural and physical environment'[1,2]. These levels are partially related. Not all impairments or functional limitations precipitate disability, and similar patterns of disability may result from different types of impairments and limitations in function[2]. Factors which may contribute to the amount of disability are the individual's definition of the situation and his reactions, the expectations and reactions of others, and the characteristics of the environment[2,9,11].

An important elaboration of Nagi's disablement concept was developed by Verbrugge and Jette[10]. They viewed disablement as the various impacts of chronic and acute conditions on the functioning of the body, human perform-

ance, and on people's functioning in usual, expected, and desired roles in society. They pointed to internal and external factors which might influence a disablement process, such as risk factors (predisposing characteristics and lifestyle), coping style, care, and special equipment and devices. Verbrugge and Jette[10p.4] defined disability as: "Difficulty doing activities in any domain of life (from hygiene to hobbies, errands to sleep) due to a health or physical problem". Disability is not a personal characteristic, but instead is a gap between personal capability and environmental demand.
Research in physical therapy aims at the dynamics and relationships along the disability spectrum[7,8]. Throughout the last decade more attention has been paid to improve a patient's function, e.g., the ability to ambulate or complete daily activities, whereas the preceding era was dominated by efforts to cure/ change impairments[12].

Disablement in childhood

The ongoing developmental process in children is one of the main reasons that disablement in childhood differs markedly from disablement in adulthood. Children grow up, learn essential functional activities, and will be able to function more and more independently, guided by the parents or caregivers. Childhood disability significantly goes beyond the impairment domain since functional ability, parental involvement, family dynamics and resources are equally important in moulding the outcome[13,14]. Disability in children can be viewed as the extent to which caregivers play a larger role in the accomplishment of daily activities than would be expected at a certain age[6]. The ultimate goal of pediatric (re)habilitation is to integrate the disabled child into the home, the school environment and the community, in which he or she learns essential functional activities independently, in a safe and timely manner. It has to be recognized that parents or caregivers participate in facilitating (new) daily activity skills[15]. This social context has to be considered when measuring a child's performance.
Haley and Coster[5] developed a disablement framework which can be applied specifically to children. They elaborated the Nagi's disablement concept

Conceptual Model of Measurement Constructs Included in the PEDI

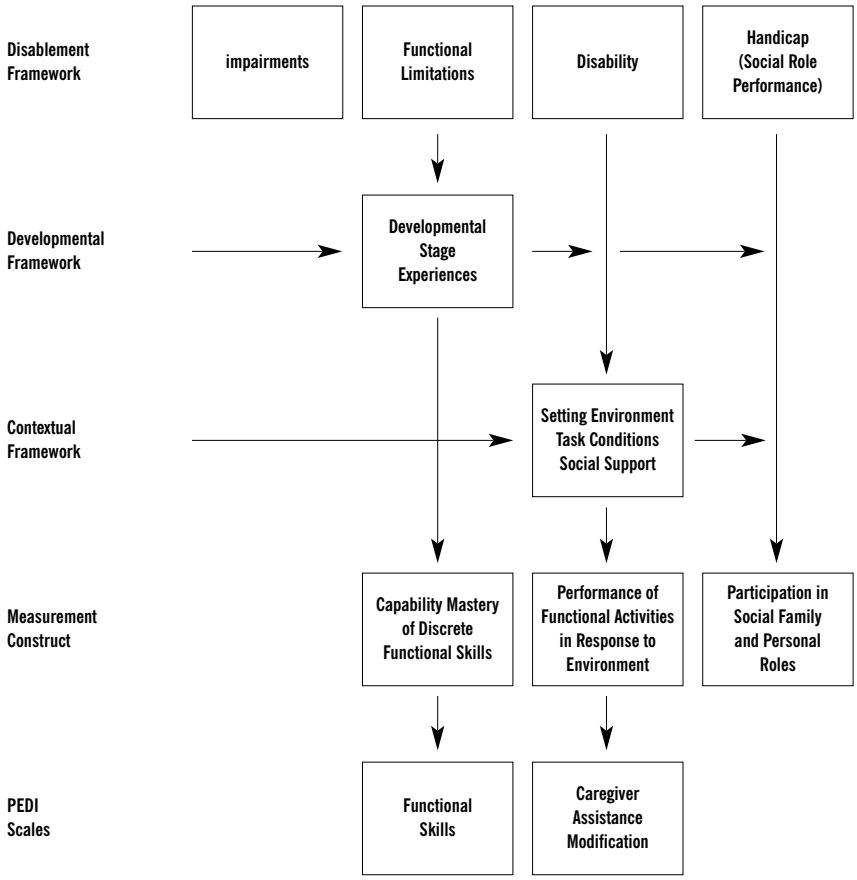

Figure 1. Conceptual Model of Measurement Constructs Included in the PEDI.

With permission reprinted from the PEDI manual[6]

incorporating a *developmental and contextual* framework, as illustrated in Figure 1. By using a standard reference it is possible to determine if the child's capabilities are age-appropriate in fulfilling essential functional activities. Additionally, contextual factors like the role of the parents, the expectations, and the availability of (re)habilitation equipment influence the degree of independency in disabled children.

Outcome measurement

Health-related outcome measures are needed to assess the different elements of the disablement process. Many different outcome measures can be distinguished and their usefulness depends on the purpose and study of interest. Generic outcome measures are instruments which are broadly applicable across diseases, and are suggested to be in favour when starting a comparative study of different diseases or populations[16]. In contrast, disease-specific outcome measures are preferred in evaluation studies when small changes in disease manifestations have to be recognized.
In addition, outcome measures can be divided into predictive, discriminative, or evaluative instruments[17].
The subject of this dissertation purports to have discriminative and evaluative potentials. Discriminative measures emphasize the ability to distinguish between individuals or groups, or when possible to compare them to an appropriate standard. They may provide a basis for diagnosis, placement, identification of delay, or determination of the level of independence[15]. Evaluative measures are able to detect longitudinal clinical change in the health status of individuals or groups. They may be used for determining treatment benefit in clinical trials and for examining the amount of functional gain in relation to intensity of services or cost of treatment[15]. Guidelines for reviewing outcome measures are still subject of interest and have been described in literature[18-21].

Pediatric Functional Status Measurement

Functional status is part of the concept of Health Related Quality of Life (HRQOL)[16]. It refers to the child's individual performance of functional daily activities. A child's functional status can be subdivided into four distinct domains: (1) physical, (2) mental, (3) emotional, and (4) social. Basic physical function items or activities of daily living (ADL) include transfers to and from a chair or bed, to dressing, feeding, and grooming[9,22]. Mental function

reflects the child's cognitive and problem solving capabilities. The term emotional function can be used to represent the child's affective and coping capabilities, whereas social function reflects the child's capability to interact appropriately with adults and peers. A child's disability may refer to an alteration in functional status in any of these four domains[22].

The concept of a functional status measure for children must yield aspects of a developmental and a contextual framework according to the childhood disablement model[5]. The child's best performance (capability, related to the child's developmental stage), as well as the actual performance (performance of functional activities in response to the environment) have to be measured to gain insight into the child's functional status. A visually impaired child in our clinic was able to manage the buttons on his clothes. Nevertheless, he always got assistance from his mother because of time constraints. The latter illustrates that the amount of assistance may indirectly reflect disablement.

Measuring the actual performance of essential activities across settings needs the use of judgement based instruments, i.e. the parents are asked to make a judgement about the ability to fulfil these activities, and more specifically, in which environmental situation (home, school, indoors, outdoors etc.) the ability was fulfilled. Hereby, the examiner relies on the perception of the parents[23].

Pediatric functional status measures differ from developmental milestone inventories, as they emphasize the degree of independency instead of the amount of developmental milestones that are mastered and they are more linked up with (re)habilitation treatment goals[15]. Furthermore, functional status measures are more focused on the quantity of essential activities (what), and are less concerned with the quality of the behavior (how)[15]. As changes in functional status of children with disabilities may be subtle, functional status measures need to have sufficient gradations to register change[15].

Pediatric Evaluation of Disability Inventory

The Pediatric Evaluation of Disability Inventory (PEDI) is a functional status instrument for infants and young children aged 6 months to 7.5 years[6]. It is a judgement-based structured interview which can be administered to parents of disabled children, or children whose functional delay is in question. The PEDI was intentionally developed to be a generic outcome measure with discriminative and evaluative potentials.

The measurement construct (see Figure 1) is based on the childhood disablement framework[5]. The capability of a child to fulfil essential functional activities can be measured using a *Functional Skills Scale* (FSS). The format of the FSS is dichotomous, i.e. questions can either have a positive or a negative score. A positive score will be given when a child has mastered the particular skill. In addition, the actual performance can be measured using a *Caregiver Assistance Scale* (CAS). The CAS determines the amount of parental assistance, which is needed for, or is given to the child. The format of the CAS is a 6-points ordinal scale ranging from 0 (= totally dependent) to 5 (= independent). Finally, *a Modifications Scale* (MS) can be used to register (specially adapted) equipment, which is normally used in performing these activities.

The PEDI measures routine functional activities in the domains: Selfcare, Mobility, and Social Function. An overview of the content is shown in Table 1 and 2, Chapter 3.

The Rasch rating scale model was used for the scale construction of the PEDI[24,25]. Furthermore, Haley et al.[6] calibrated the PEDI using this model and a normative sample of 412 non-disabled American infants and young children. For the functional skills scale this calibration rendered an ordering of the items according to difficulty. For each outcome scale, the calibration also rendered a normative distribution of the summed scores (the number of positive responses per outcome scale) in 14 age-classes (14 six-months intervals from 0.5 years through 7.5 years). These normative distributions were subsequently standardized for each age-class (mean score: 50; standard deviation: 10). The score of an individual child can be assessed against this standard to determine if that child has a functional status typically seen for that age.

In addition, summed scores were transformed into scaled scores, which provide an indication of the performance of the child along the continuum of relatively easy to relatively difficult items. Scaled scores are distributed along a scale from 0 to 100, with increasing numbers representing increasing degrees of functional performance. Scaled scores are not adjusted for age and therefore can be used to describe the functional status of older children with disabled conditions.
Finally, the Rasch rating scale methodology enables the researcher to perform goodness-of-fit analysis between individual child profiles and the hierarchical model in order to identify children with unusual performance patterns.
The PEDI is internationally recognized as a useful outcome measure[26-35]. Research activities with the PEDI in the Netherlands were performed in children with several chronic conditions[36-38]. Based on these studies and supported by scientific literature concerning cross-cultural adaptation of outcome measures[24,39-41], a discussion was started whether or not the PEDI is applicable for use in the Netherlands.

Aim of the study

The aims of this thesis are:
- to investigate the applicability of the PEDI in the Netherlands
- to investigate the content validity of the Dutch adapted PEDI.
- to investigate the reliability of the Dutch adapted PEDI.
- to investigate the discriminative validity of the Dutch adapted PEDI.
- to investigate the responsiveness of the Dutch adapted PEDI.

Outline of the thesis

Chapter 1 is a brief introduction of conceptual issues regarding childhood disablement and pediatric functional status measures, as well as an introduction of the conceptual model and measurement construct of the PEDI.
Chapter 2 deals with the applicability of the PEDI in Dutch children, and discusses the pitfalls when using the PEDI in other than the target population. The Dutch adaptation process and a content validity study are described in Chapter 3. Different kinds of reliability of the Dutch adapted PEDI are tested and results are presented in Chapter 4. Chapter 5 deals with the discriminative validity of the Dutch PEDI when used in a combined clinical sample of different pediatric diseases and disorders. In chapter 6, the responsiveness of the Dutch PEDI was examined using an alternative approach in responsiveness studies. Chapter 7 contains a summary and general discussion.

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Cultural Differences in Functional Status Measurement

-Analyses of Person Fit According to the Rasch Model-

Chapter 2

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Abstract

For many reasons it is preferable to use established health related outcome instruments. The validity of an instrument however, can be affected when used in another culture or language other than what is was originally developed. In this paper, the outcome on functional status measurement using a preliminary version of the Dutch translated 'Pediatric Evaluation of Disability Inventory' (PEDI) was studied involving a sample of 20 non-disabled Dutch children and American peers, to see if a cross-cultural validation procedure is needed before using the instrument in the Netherlands. The Rasch model was used to analyze the Dutch data. Score profiles were not found to be compatible with the score profiles of American children. In particular, ten items were scored differently with strong indications that these were based on inter-cultural differences. Based on our study, it is argued that cross-cultural validation of the PEDI is necessary before using the instrument in the Netherlands.

Introduction

At the moment there is increasing interest in health outcome assessment, not only in terms of biomedical parameters, but more so in quality of life and health status. This kind of assessment may be utilized for evaluation purposes of interventions, as well as health policy makers with regard to effectiveness of health care programs, epidemiological studies and clinical trials[1]. In general, the development of outcome instruments is a time consuming process. Besides this, from a research point of view, a proliferation of many specific instruments for the same condition threatens to decrease the external validity of study findings[2]. Therefore, it is preferable to use established instruments. The validity of an assessment however, can be affected when an outcome instrument, developed and calibrated for a specific culture, is used in another culture[1,3-6], due to translation difficulties, irrelevancy of item contents, and inappropriate norm scores.

In this paper we present the difference in outcome on functional status measurement between a sample of non-disabled Dutch children and American peers. The purpose was to study if a cross-cultural validation procedure of the 'Pediatric Evaluation of Disability Inventory' (PEDI)[7] is needed before using the instrument in the Netherlands. The Rasch model was used to analyze the score profiles with respect to the functional skills of 20 children. See Fischer and Molenaar[8] for a comprehensive discussion of the Rasch model; Wright and Stone[9] and Wright and Masters[10] for introductions. Furthermore, see Molenaar and Hoijtink[11], and Klauwer[12] for Rasch model based analysis of score profiles, better known as person fit analysis.

It will be illustrated in this paper, that the score profiles of the Dutch children, on one of the functional skill domains (social functioning) were not compatible with the score profiles of American children. These findings will be discussed in relation to recent literature. Based on these findings, it will be argued that cross-cultural validation of the PEDI is necessary.

PEDI

The PEDI is a parental-report, or structured-interview, instrument used by pediatric physical therapists and other rehabilitation professionals to assess functional abilities of young children. This reference-based instrument, developed in the USA, is intended to be used to discriminate between non-disabled and disabled children, and to guide and evaluate pediatric rehabilitation programs. The PEDI scales consist of a 'functional skills scale' and a 'caregiver assistance scale'[13]. The functional skills scale comprises questions concerning routine daily activities in childhood divided into three domains: Selfcare (73 questions), mobility (59 questions), and social functioning (65 questions). The format is dichotomous, i.e., questions can be scored either positive or negative. A positive score will be given when a child has mastered the particular skill.

The caregiver assistance scale comprises 20 questions concerning the same activities. Using six rank-ordered response choices, ranging from 0 (= totally dependent) to 5 (= independent), the amount of assistance could be scored. Every item has its own score criteria described in the PEDI manual. Scores 0 and 1 refer to the participation of the caregiver in more than half of the activities, while score 2 to 5 refer to a progressive independence of the child.

These two scales consist of three domains, selfcare, mobility, and social functioning -, resulting in six outcome scales. The selfcare domain refers to activities of eating, grooming, bathing, dressing, and bladder/bowel management. The mobility domain refers to transfer activities at home and in the community setting. The social function domain refers to communication, social interaction, orientation of time and place, and household and community function.

Validation and reliability studies of the PEDI were executed. Inter-interviewer reliability was good to excellent (Intra Class Correlation (ICC) coefficients ≥ 0.96[7], ICC ≥ 0.85[14]). Intra-interviewer reliability varied from moderate to excellent (ICC ≥ 0.80[14], ICC ≥ 0.67[15]), while varying degrees of inter-respondent (parent vs. health care professional) reliability (ICC 0.18 - 0.94[15], ICC ≥ 0.84[7]) were observed. Content validity[16] and concurrent and construct validity[17] were viewed as acceptable at the least. These studies were executed in the USA and Canada.

Calibration of the PEDI using the Rasch model

Haley[7,18] calibrated the PEDI using the Rasch model, and a normative sample of 412 non-disabled American infants and young children, aged 0.5-7.5 years, divided into 14 age-classes. This calibration rendered for the functional skills scale had an ordering of the items according to difficulty (is denoted by the Greek symbol δ). The social function skills scale for example, has a range of difficulty from δ = -19.36 to 20.24. Item 29: " Does your child attempt to imitate adult's previous action during a play activity?" which is rather easy, had a δ of -2.02. Item 33: "Does your child tries to work out simple plans for a play activity with another child?" is rather difficult, which is expressed by a δ of 2.04.

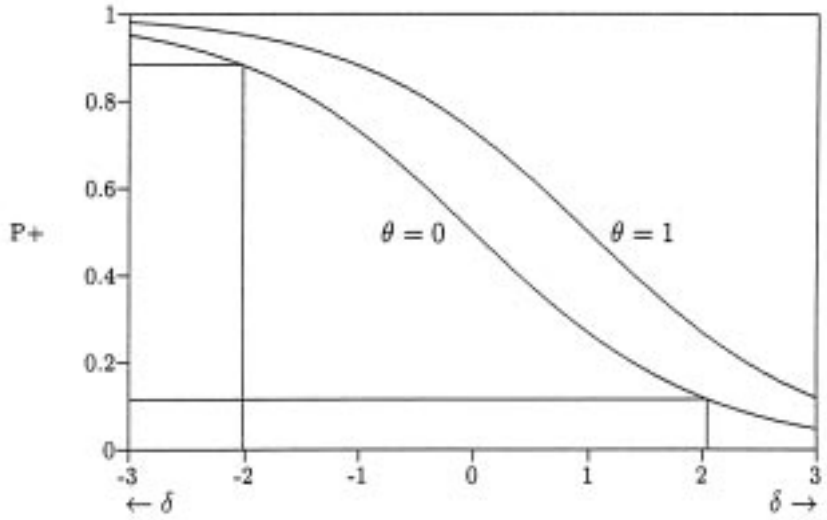
For each outcome scale, the calibration also rendered a normative distribution of the summed scores (i.e. the number of positive responses per outcome scale) for each age-class. These normative distributions are standardised for each age-class, resulting in a mean of 50 and a standard deviation of 10. By comparing a standardised score of an individual child with his/her peers from the normative sample, we are able to judge if that child has a functional status typically seen for that age. Accordingly, the PEDI can be used to discriminate between non-disabled and disabled children.

As indicated above, in the Rasch model each item is characterised by its difficulty using δ. Similarly, for each outcome scale, each person is characterised by his or her ability (to be denoted by θ). This is another transformation (like the standardised score) of the summed score for an outcome scale.

The relation between δ, θ and the probability of a positive response p+ (i.e. the child possesses the skill at hand) is presented in Figure 1 for a child with $\theta = 0$ and $\theta = 1$. One notices, the more difficult the item (i.e. skill), the lower the probability of skill possession. Such a profile can be calculated for each child. It is the basis of a goodness of fit test that can be used to investigate if a child's response pattern is deviant or not. For example, a child who did not master some of the easy skills, but did master some of the more difficult skills could be classified as being deviant. As shown in Figure 1, in the child with $\theta = 0$, it would be unusual not to have mastered a skill with diffi-

culty δ = -2.02 (p+ = 0.88). Similarly, it would also be unusual to have mastered a difficult skill δ = 2.04 (p+ = 0.12). The Data entry program for the PEDI, as developed by Haley[7], computes a goodness of fit test, that summarises, for each child and each outcome scale, the differences between the observed scores and the corresponding scores p+[10]. For the American calibration sample, most of the inferred fit scores had values between -2 and +2, except for the fit scores on the social skills scale, of which 15% were above +2[7].

Figure 1. Relation between item difficulty δ, person ability θ, and the probability of a positive response p+.

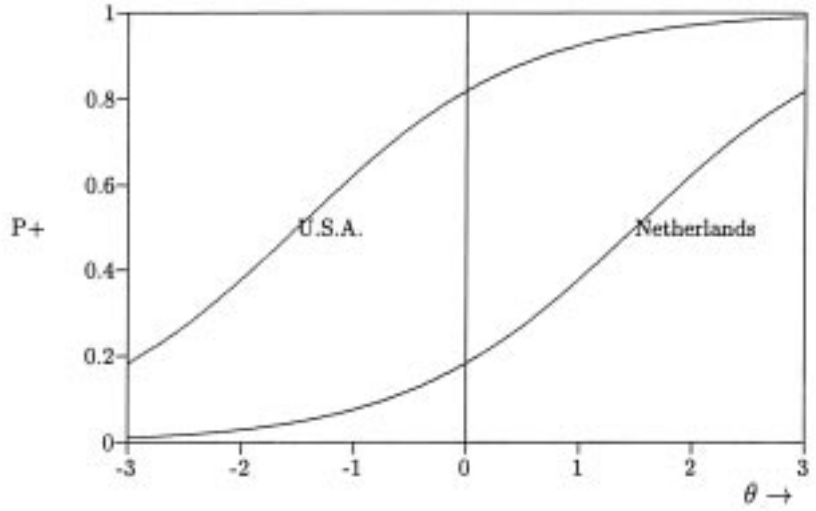


Using the Rasch model for cross-cultural validation

The Rasch model can be used for cross-cultural validation of the PEDI. In the standard approach the questions of the PEDI have to be scored for about 200 children from the United States and the Netherlands. Subsequently, the PEDI should be calibrated (i.e. the difficulty is computed for each question) separately for the samples from the US and the Netherlands. If the difficulties of the items are the same in both the countries (see Andersen[19] and Glas and Verhelst[20] for two different approaches towards testing the hypothesis of no cultural differences) there are, according to the Rasch model, no cross-cultural differences. The consequence of cultural differences is illustrated in Figure 2.

The line labelled "USA" represents the probability of a positive response to a certain item in the USA for different ability levels. The line labelled "Netherlands" represents the probabilities for the same item, but now in the Netherlands. As can be seen, for all ability levels, the probability of a posi-

Figure 2. Relation between person ability θ and the probability of a positive response p+ for the same item, for children from the USA and children from the Netherlands.



tive response is larger in the USA but the same item is more difficult for children in the Netherlands. If such a biased question is used in a questionnaire, the ability of children in the Netherlands would be underestimated - compared to those in the USA. Stated otherwise, use of such a question implies the introduction of cross-cultural bias.

In our project, for each item, only the line labelled "USA" can be constructed. The sample of 20 children from the Netherlands is not sufficient for a reliable construction of the line labelled "Netherlands". Stated otherwise, the standard approach for investigating cross-cultural validation cannot be applied here. An alternative approach is implied in the paper by Molenaar and Hoijtink[21].

As indicated in the previous section, for each child from the Netherlands a fit statistic could be computed which will indicate if the responses were consistent with the item difficulties obtained from the calibration sample from the USA. If, most of the children had such inconsistencies, this implies that the "USA calibration" does not give a good description of the item - difficulties in the Netherlands i.e. inconsistencies imply cross-cultural differences. Subsequently, for each child and each item the response is compared with the corresponding p+, an indication is obtained about the items causing the inconsistencies. Inconsistencies arise (see also the formula of the fit statistic) if p+ is large (say larger than 0.75) and a child responds to 0, or if p+ is small (say smaller than 0.25) and a child responds to 1. If five or more of these inconsistencies are found for the same item, this is an indication that the item is more difficult or less difficult in the USA than in the Netherlands. Stated otherwise, five or more of these inconsistencies for the same item point to a culturally biased item.

Methods

Subjects

The Dutch sample comprised of 20 children under the age of 7½ years (♂:♀ = 8:12; mean age: 56 months; age range: 23-86). They were recruited as controls for a study in children with cerebral palsy (CP)[22]. Parents of children who attended a regular school or pre-school facility were asked to participate. Only non-disabled children were included in this control group. A selection was made to match this group with the CP group for age and gender. For our study we had permission to use the raw scores of the controls for analysing their profiles.

Instrument

Functional status was measured with a preliminary version of a Dutch translation of the PEDI. The translation was performed by a multidisciplinary collaborating study group, consisting of professionals active in the domain of clinical rehabilitation. Two interviewers followed a training procedure before starting the assessment. Inter-interviewer reliability of the translated version was studied. The ICC varied from 0.8 to 1.0[22]. Level of functional status was calculated by summing raw data into summed scores and transforming these into normative standard scores, according to the guidelines in the PEDI manual[7]. The results on each outcome scale was compared with the reference group in the USA (normative standard score USA: $\bar{x} = 50 \pm 10$)[7]. The results are shown in Table 1. Fit scores were calculated using the Data entry program, software specifically developed for the PEDI[7]. For each outcome scale, the fit scores of the 20 Dutch children were compared with the fit scores of the American calibration sample (normal fit-scores USA: $-2 \leq x \leq +2$)[7]. The results are presented in Table 1. If deviant fit scores were found, the score profiles were analysed to determine which items were responsible for the lack of fit. Therefore, the observed scores were compared with p+ per item for each child.

Statistics

Normative standard scores reflecting the level of functional status, and fit scores which reflect the item response patterns, are expressed as medians (P50) and quartile values (P25-P75) for each outcome scale. In case of deviant fit scores in a particular scale, the number of children who scored a large difference between p+ and the observed score was counted per item.

Results

In Table 1, the median and quartile values of the normative standard scores are presented. For example, the median of the selfcare scale on the functional skills was 45.0 and the quartile values were 38.3 and 54.1, respectively. Presented this way, it looks as if the normative standard scores of the non-disabled Dutch sample are within normal proportions, from which one might conclude that their level of functional skills and caregiver assistance on the three domains could be considered as typical for that age.
However, the fit-scores of the social function scale, functional skills, are deviated when considering a fit score between -2.0 and +2.0 as a reflection of a score profile, which was expected (Table 1). The median on this scale was +3.0, and more than 75% (17/20) of the fit scores on this scale was above +2.0.

Table 1. Functional status in non-disabled Dutch children (n=20)

PEDI scales	Normative standard scores		Fit scores	
	P50	P25-75	P50	P25-75
Functional skills				
Selfcare	45.0	38.3 - 54.1	0.7	0.2 - 1.8
Mobility	43.3	38.4 - 51.2	0.3	-0.5 - 0.7
Social function	48.2	44.5 - 62.4	3.0	2.3 - 3.4
Caregiver assistance				
Selfcare	41.2	35.2 - 45.0	-0.3	-0.7 - 0.2
Mobility	44.7	38.2 - 51.9	0.1	-0.4 - 0.9
Social function	49.1	45.1 - 56.4	-0.3	-0.9 - 0.0

Based on the American calibration sample[7]:
Normative standard scores: $\bar{x} = 50 \pm 10$; normal range 30-70.
Fit-scores: $-2 \leq x \leq +2$.

Table 2. Fit score analysis

Child	Item 35 [a]		Item 58 [a]		Item 45 [a]		Item 64 [a]		Item 33 [b]		Item 47 [b]		Item 5 [c]		Item 14 [c]	
	p+	Obs	p+	Obs	p+	Obs	p+	Obs	p+	Obs	p+	Obs	p+	Obs	p+	Obs
1	0.0121	0	0.0094	0	0.0002	0	10^{-5}	0	0.030	0	0.014	0	0.002	0	0.008	0
2	0.9432	1	0.9282	1	**0.2869**	**1**	**0.0218**	**1**	0.999	1	0.998	1	0.144	0	0.410	0
3	0.2091	0	0.1707	0	0.0063	0	0.0003	0	0.180	0	0.080	0	0.031	0	0.117	0
4	0.9994	1	0.9992	1	0.9774	1	0.7068	1	0.996	1	0.991	1	0.790	1	0.940	1
5	0.8839	1	0.8556	1	**0.1557**	**1**	**0.0101**	**1**	0.968	1	0.924	1	0.579	1	0.850	1
6	0.8053	1	0.7631	1	**0.0911**	**1**	**0.0055**	**1**	0.991	1	0.979	1	**0.093**	**1**	**0.299**	**1**
7	0.8839	1	0.8556	1	**0.1557**	**1**	**0.0101**	**1**	0.999	1	0.998	1	**0.093**	**1**	**0.299**	**1**
8	0.6177	1	0.5572	1	0.0376	0	**0.0021**	**1**	0.078	0	**0.033**	**1**	0.043	0	0.157	0
9	0.9797	1	0.9741	1	0.5399	1	**0.0612**	**1**	0.977	1	0.945	1	**0.641**	**0**	0.698	1
10	0.9994	1	0.9992	1	0.9774	1	0.7068	1	0.996	1	0.991	1	0.797	1	0.942	1
11	0.9432	1	0.9282	1	**0.2869**	**1**	**0.0218**	**1**	0.957	1	0.900	1	0.579	0	0.850	1
12	0.9958	1	0.9946	1	0.8532	1	**0.2441**	**1**	0.944	1	0.873	1	0.093	0	0.299	0
13	0.9797	1	0.9741	1	0.5399	1	**0.0612**	**1**	0.999	1	0.998	1	0.579	0	0.850	1
14	0.9797	1	0.9741	1	0.5399	1	**0.0612**	**1**	0.846	1	**0.689**	**0**	0.214	0	0.529	0
15	0.7170	1	0.6637	1	**0.0578**	**1**	**0.0034**	**1**	0.846	1	0.689	1	0.144	0	0.410	0
16	0.9999	1	0.9999	1	0.9986	1	0.9761	1	0.985	1	0.963	1	0.144	0	0.410	0
17	0.0469	0	0.0369	0	0.0011	0	**6×10^{-5}**	**1**	**0.257**	**1**	0.122	0	0.013	0	0.054	0
18	0.4255	0	**0.3658**	**1**	**0.0176**	**1**	**0.0009**	**1**	**0.681**	**0**	0.462	0	0.020	0	**0.077**	**1**
19	0.9797	1	0.9741	1	0.5399	0	**0.0612**	**1**	0.894	1	0.773	1	0.214	0	0.529	1
20	0.9999	1	0.9999	1	0.9999	1	0.9987	1	0.985	1	0.963	1	0.797	1	0.942	1

[a] Item number from social functioning skills scale. [b] Item number from selfcare skills scale. [c] Item number from mobility skills scale.

p^+ = expected score; obs = observed score.

In Table 2, four examples of the social function skills are shown highlighting the differences between the expected score (p+) and the observed score per child. Items 35 and 58 are scored as was expected by nearly all children, while items 45 and 64 obviously are not. As one could notice for item 64, the observed scores of most of the children (18/20) are 1, including 14 children with large differences between the observed scores and p+. Table 2 contains two examples of both the selfcare domain as well as the mobility domain. Most of the observed scores were compatible with the calculated scores.

In Table 3, we present a frequency table of items on the social function skills scale, which were scored unexpectedly by 5 or more children. The number of five children (25%) was, in our opinion, large enough to include these items in our analysis. The first column refers to the item number of the PEDI. For example, item 59 was scored unexpectedly by 9 children (9/20). All these children have an observed score of 0 with large differences compared to p+. In conclusion, 6 items (item number 40, 45, 49, 55, 59, and 64, respectively) were strongly marked because eight or more children scored unexpectedly, while four items (item number 30, 34, 54, and 65, respectively) were scored unexpectedly by five to seven children.

Table 3. Frequency table of unexpected scores social function skills scale

Item number [a] PEDI	**Frequency** [a] (n=20)	**Score = 0** (score = 1 expected)	**Score = 1** (score = 0 expected)
30	5	5	0
34	5	5	0
40	10	10	0
45	11	1	10
49	10	8	2
54	5	3	2
55	8	6	2
59	9	9	0
64	14	0	14
65	6	0	6

[a] Only items with unexpected results in 5 or more children are presented in this table.

Discussion

The purpose of our study was to determine if a cross-cultural validation procedure of the PEDI is needed, before using the instrument in the Netherlands, by comparing the outcome of non-disabled Dutch children with American peers. Regarding the size of the normative standard scores of the three domains, it appeared as if the Dutch sample did not have a specific outcome on face value, although, based on the results, one may have the impression that they have slightly lesser skills. Nevertheless, for the social skills scale, almost all (17/20) fit values are larger than +2, i.e. larger than most of the values observed in the American calibration sample. Since the 20 children were "normal" children, this lack of fit scores is surprising.

In two clinical samples[23,24], we found deviant fit scores on the same scale (i.e. social function). In these studies, each performed by different interviewers, we included 41 children totally. Most of these children were known for their physical disabilities in daily life, but more importantly, their social functioning was not affected. While we used different, trained interviewers in both the non-disabled group as in the clinical sample, it is unlikely that the deviant fit scores were due to misinterpretations of the interviewers or respondents during their interviews[18].

A plausible explanation could be the existence of cultural differences between the American and Dutch children. This would imply that the calibration and subsequent calculation of norm scores based on the American children cannot be used for the Dutch children in the social function domain. To find support for this assumption, the score profile of each child was analyzed, to determine, which items were responsible for the lack of fit scores. If 'responsible' items could be found, and, an interpretation could be given, culturally incomparability could be identified, and this would justify a calibration of the PEDI using a Dutch calibration sample; if the instrument were to be used in Dutch children. 'Responsible' items are items where the observed score is rather different from p+, the probability of a positive response.

Based on our analyses using the Rasch model, we found 10 PEDI items in the social function skills scale (Table 3, appendix) which were thought to be responsible for the deviant fit scores, and for which we can give an interpre-

tation. The selfcare and mobility scales were not analyzed in detail as was done for the social skills scale, because these scales had no overall deviant fit scores.

According to Wright and Masters[10] if the values of the person fit statistic is larger than 2, a lack of fit is indicated. The number 2 corresponds (approximately) with an error of 0.025. The fit statistic investigates whether the responses of a Dutch child are consistent with the item difficulties calibrated, using the sample from the US. Under the null-hypothesis, none of the 20 children have inconsistent response patterns, the expected number of children with a fit statistic larger than 2 is approximately 20 x 0.025 = 0.50. For social functioning 17 children have a fit statistic larger than +2. Since 17 is substantially more than 0.50, this is a clear indication that the rating of item difficulty as calibrated in the US does not correspond with the item difficulties in the Netherlands.

For selfcare and mobility 1 and 0 children, respectively, had a fit statistic larger than +2. The number of children in these domains with a fit statistic larger than +2 is nearly equal to the expected number of children (0.50) with a fit statistic larger than +2, if all response patterns are consistent with the item difficulties as calibrated in the US, i.e. the set of items appear to be cross-culturally comparable. Note that this does not imply that none of the individual items are culturally biased. The latter is hard to investigate using only 20 persons. To do this, we planned extended studies in future by means of DIF analysis.

One of the problems using health related outcome instruments cross-culturally could be that the item contents are not equivalent regarding relevancy[1]. The particular items which were scored differently, however, are basic social skills throughout the world and therefore relevant in both cultures. More appropriate is the explanation that the sequence of development of these social skills could be different or culture specific. As one can notice in Table 3, US children seem to develop play- and communication based skills (PEDI items 30, 34, 40, 49, 59), and household chores (PEDI item 55) at an earlier phase than Dutch children. However, Dutch children develop community skills (PEDI items 45, 64, 65) at an earlier phase. It is beyond the scope of this study to reveal the causes of these differences; different traditions in upbringing as well as the influence of the school system and the way society is arranged could all contribute to the differences found in the study.

Note that our analysis is descriptive since our sample was small. The conclusions are also based on a study with a preliminary translated version of the PEDI. Theoretically, the deviant fit scores could also be forthcoming from translation problems. Nevertheless, it is remarkable that we found many deviant fit scores in many items of the social skills scale, and the differences in outcome are perceived as cultural differences in the upbringing of children. The purpose of our study was to investigate the possible presence of inter-cultural differences, which would require a re-validation procedure and not to investigate inter-cultural differences in detail. In that case, the Rasch model has to be used in the standard approach[19,20]. We acknowledge the fact that these results should be validated on larger samples when comparing item difficulties across cultures.

Finally, the consequences of inter-cultural differences can not only influence the response patterns, but also affect the normative standard scores. Hypothesized, that all the Dutch children scored zero in a particular item where a score 1 was expected. Based on cultural differences, the consequences would be that the score of the Dutch children would be underestimated, while reference values based on the American calibration sample cannot be simply applied.

In conclusion, the indications of the existence of inter-cultural differences based on our study findings, the plausible influence on outcome results and the pitfalls as described in the reference[1,3-6], in using health related outcome instruments cross-culturally, are strong arguments to validate the PEDI before using the instrument in the Netherlands. At this moment a new translation by an independent and certified translator, an adaptation based on 'content equivalents', and a re- translation procedure by an independent native speaker is almost completed.

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Appendix Chapter 2

Content of relevant PEDI items on the social functional skills scale

item 30 During play child may suggest new or different steps, or respond to adult suggestion with another idea (During block building child says, "Let's make a door now." Adult starts to build, child says "no, how 'bout here?")

item 34 Plans and carries out co-operative activity with other children; play is sustained and complex (Makes up and acts out a story with another child; works with a peer to build a castle from blocks)

item 40 Makes up elaborate pretended sequences from imagination (Makes up and carries out stories with pretended characters - monsters, princesses, etc., not just acting out an exact story from a book or television show, or own experience)

item 45 Can direct an adult to help child return home or back to the hospital room (When in a car, child can show adult the route from school to home; if in hospital, child can point to correct floor button and show which direction to room)

item 49 Associates a specific time with actions/events (Knows time of favorite television show)

item 54 Occasionally initiates simple household chores; may require physical help or reminders to complete (Child asks to set the table, and may need a reminder about where things go)

item 55 Consistently initiates and carries out at least one household task involving several steps and decisions; may require physical help (Child gets pile of own clean clothes and puts them away in drawers)

item 59 Knows not to accept rides, food or money from strangers

item 64 Explores and functions in familiar community settings without supervision: neighborhood, hospital, or school areas (Child can follow directions to the school office and return. Can explore and function in neighborhood for several blocks; doesn't get lost.)

item 65 Makes transaction in neighborhood store without assistance (Child can go into drugstore without an adult and buy gum)

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Dutch adaptation and content validity of the 'Pediatric Evaluation of Disability Inventory' (PEDI)

Chapter 3

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Abstract

Purpose: To adapt the American PEDI into a Dutch version, and to establish the content validity of this pediatric functional status instrument.
Methods: The adaptation process was based on current scientific guidelines in the field of cross-cultural research. Thirty-one allied health professionals completed a validity questionnaire for the content validity study.
Results: The topic 'bicycling' was added to the questionnaire, and adaptations of the text were made without losing the content of the original PEDI. At least 81% of the respondents rated that the most important facets in examining functional status in childhood were represented in the Dutch PEDI. Eighty seven percent and 71% of the respondents rated that the PEDI is feasible for discriminative purposes and evaluative purposes, respectively.
Conclusion: The expert panel confirmed the functional content and feasibility of the Dutch PEDI for pediatric rehabilitation outcome measurement.

Introduction

An era of assessment and accountability in medicine has provided health scientists with a wide variety of health related outcome instruments[1]. Patrick and Deyo[2] warned that too many instruments for the same condition threaten the external validity of study findings. Moreover, the need for international collaboration that grew from limited (financial) resources in health research encouraged scientists to adapt already existing health outcome instruments to their needs, while maintaining good psychometric properties. Cross-cultural adaptation of existing outcome instruments evolved from a growing body of knowledge that recognized cultural differences between distinct concepts of health in different populations. It became apparent that literal translation of existing health outcome questionnaires lacked validity[2-9].

The first aim of this study was to adapt the Pediatric Evaluation of Disability Inventory (PEDI)[10], a functional status instrument for infants and young children, for the Dutch population following current guidelines for cross-cultural adaptation[4,6]. Therefore, the criteria of the European Research Group on Health Outcomes (ERGHO) that are content equivalence, semantic equivalence, and conceptual equivalence[6] were applied.

The second aim of this study was to perform a content validity study to confirm the functional content of the adapted Dutch PEDI. The adaptation process and the adaptations made as well as the results of the content validity study are reported in this paper. In future, additional reliability studies and validity studies have to complete the cross-cultural validation procedure. It will provide an outcome instrument feasible for use in a relatively small language domain, at costs far below the costs of developing a complete new outcome measure.

Cross-cultural adaptation

Cross-cultural research aims at the development of outcome instruments to measure the same phenomenon in different cultures, and to perform outcome studies of patient populations across cultures. Berry et al.[11] described

three basic approaches in cross-cultural research. The first is the 'absolutist' approach, which assumes that concepts being measured will be largely invariant across cultures. Those who only translate an instrument rather than investigate the relevance of the concepts are following this approach. The second is the 'universalist' approach, which assumes that a context-free definition and measurement of concepts will be difficult or impossible to achieve, and that measurement in a cross-cultural context will require the need for adapted instruments. The third is the 'relativist' approach, which assumes that it is impossible to use standard instruments across cultures because the variation of the culture's behavior is substantial. Adaptation of outcome measures mainly depends on the approach one favors.

The European Research Group on Health Outcomes (ERGHO) sets a minimum of criteria for cross-cultural adaptation[6]. These criteria are: content equivalence, together with semantic and conceptual equivalence. Content equivalence refers to the observation whether the concept of each item is relevant to the cultural setting. Semantic equivalence is aimed at the emphasis that must be placed on retaining the essence of what is being asked or stated rather than obtaining a direct (literal) translation of the words. Finally, conceptual equivalence is when the instrument is found to be measuring the same concept in different cultures. This can be measured by comparing the outcome on specific study populations in the original and target culture.

Herdman et al.[7] reviewed definitions of equivalence in cross-cultural research, and found little similarity and clarity, especially in 'conceptual equivalence'. The interpretation of 'conceptual equivalence' varied, between authors, from similarity of the underlying theoretical concept to similarity of the meaning of the items and similar ranking of the items of a scale. Herdman therefore strongly suggested using standardized terminology in the cross-cultural adaptation of outcome instruments. Conceptual equivalence, in Herdman's opinion, does not exist at item level, but at a higher, more abstract level, that of the way a concept is organized and expressed in different cultures[7].

To meet the criteria of content-, and semantic equivalence, Guillemin et al.[4] stressed the importance of several forward and back translations performed by qualified, independent, and bilingual translators. These translators should preferably translate into their mother tongue. The differences identified

should be addressed and when necessary the original developer should be contacted. Many researchers in this field[3,4,6] stated that a review committee should compare original and translated versions regarding content and construct of the questionnaire. Such a committee is also likely to modify or eliminate irrelevant, inadequate and ambiguous items and may generate substitutes better fitting the cultural target situation, while maintaining the general concept. After the instrument is modified by the review committee pretesting has to be started. Analysis of the responses of the pretesting phase will identify problems in the interpretation of the content. It also reveals if questions give rise to reluctance or hesitation.

PEDI

The PEDI, developed by Haley et al.[10] is a clinical instrument for the assessment of functional status in children up to 7.5 years of age. It is a judgement based parent structured interview used by professionals in rehabilitation medicine. The PEDI is able to measure both *capability* (what the child can do) and *performance* (what the child actually does do) of routine daily childhood activities in the selfcare, mobility, and social function domain, as showed in table 1.

Capability of a child can be measured using the three *functional skills scales* of the PEDI. These scales contain a total of 197 questions. The questions are supported by an explanatory part. Performance of a child can be measured using the three *caregiver assistance scales* and *modifications scales* of the PEDI[10]. The caregiver assistance scales contain 20 questions concerning the same activities of the functional skills scales. The modification scales are measures of environmental modifications and equipment used by the child in routine daily activities. The construct of the PEDI is presented in table 2.

Although the PEDI is primarily designed for functional evaluation of young children, the PEDI can also be used for the evaluation of older children if their functional abilities fall below that of children up through the age of 7.5 years. The PEDI is an appropriate instrument for measuring functional status in children with physical or both physical and cognitive disabilities. It is a less suitable instrument for assessing infants less than one year of age and

Table 1. Item content of the PEDI (USA version).

Functional skills		
Self-Care Domain	**Mobility Domain**	**Social Function Domain**
Types of food textures	Toilet transfers	Comprehension of word meanings
Use of utensils	Chair/wheelchair transfers	Comprehension of sentence complexity
Use of drinking containers	Car transfers	Functional use of communication
Tooth brushing	Bed mobility/transfers	Complexity of expressive communication
Hair brushing	Tub transfers	Problem-resolution
Nose Care	Indoor locomotion- methods	Social interactive play
Hand washing	Distance/speed indoors	Peer interactions
Washing body and face	Pulls/carriers objects	Play with objects
Pullover/front-opening garments	Outdoor locomotion- methods	Self information
Fasteners	Distance/speed outdoors	Time orientation
Pants	Outdoor surfaces	Household chores
Shoes/socks	Upstairs	Self protection
Toileting tasks	Downstairs	Community function
Management of bladder		
Management of bowel		
Subscales per domain		

Table 2. Construct of the PEDI (USA version)

	PEDI scales		
	Capability	Performance	
PEDI domains	Functional skill scale	Caregiver assistance scale	Modifications scale
Selfcare	73	8	8
Mobility	59	7	7
Social Function	65	5	5
Format	Dichotomous	6-point ordinal scale	4-point ordinal scale

Domains, scales and number of items in the PEDI

for assessing older children with minimal disability. Its validity for children whose primary disability is behavioral or social, whose functional performance shows significant fluctuations, or whose functional limitations are thought to be in the mild to moderate range is still under investigation[10]. The PEDI is intended to be used to discriminate between non-disabled and disabled children, and to guide and evaluate pediatric rehabilitation programs. Extended reliability and validity studies with the original PEDI were published[10,12 -16]. Haley et al.[17] performed a content validity study in the developing phase of the PEDI. This study provided information whether the items of the PEDI, both individually and as a whole, represent the construct that it was supposed to measure. The respondents who participated in that study confirmed the accuracy of the content of the PEDI.

Dutch adaptation process of the PEDI

In line with ERGHO, the 'universalist' approach[11] was adopted, which assumes that measurement in a cross-cultural context requires the use of adapted instruments. The main argument was based on the findings of a preliminary study of Custers et al.[18] in which the responses on the PEDI of 20

healthy Dutch children were investigated and compared to American peers. In this study, person fit was analyzed according to the Rasch model[19,20]. Score profiles of the Dutch children were found to be incompatible with the score profiles of American peers indicating cultural differences. It was agreed upon that a cross-cultural adaptation procedure of the PEDI was inevitable.

To obtain content, semantic, and conceptual equivalence[6], the Dutch adaptation process of the PEDI was based on the guidelines for cross-cultural adaptation of Herdman et al.[7], Bullinger et al.[3], Guillemin et al.[4] and Touw-Otten and Meadows[6], meeting the criteria of the ERGHO. In accordance with Herdman, the interest was in whether the underlying concept of the PEDI was appropriate to use in the Netherlands.

The theoretical concept of the PEDI is based on the disablement model of Nagi[21]. Two concepts of this model, 'functional limitations' and 'disability', are implemented in the PEDI[10,22]. Current modifications of this model were made by Jette & Verbruggen[23], and the US National Center for Medical Rehabilitation Research (NCMRR)[24]. At the moment, it is still a leading model in health related outcome studies in the USA. Recently, this model was also used in Swedish and Dutch health related outcome studies[25-27]. Moreover, the latest draft of the WHO's 'International Classification of Impairments, Disabilities, and Handicaps' (ICIDH2-B2)[28], although still undergoing evaluation, shows more and more similarity with the modified disablement model of Nagi. As the underlying concept of disability, i.e. limitations of daily activities, do not differ significantly, it is felt that the underlying concept of the American PEDI is appropriate to use in the Netherlands. Therefore, the 'relativist' approach[11], which assumes that the PEDI is not feasible to use at all, was rejected.

A certified translator translated the PEDI into the Dutch language. She was previously informed about the PEDI, the aim of the translation, and the criteria stated by Guillemin et al.[4]. The Dutch translated version was then subjected to an adaptation process. A multidisciplinary review committee compared the original and translated version. The members of this committee were professionals in the field of pediatric rehabilitation (n=4) and researchers in educational sciences (n=2). This committee investigated 'content equivalence'and 'semantic equivalence' by reviewing and adapting the PEDI items attentively. They also investigated additionally the accuracy

of the items in terms of ambiguity and completeness, and the construct of the items in terms of redundancy of text.

Although, it was intended to change the PEDI only if necessary, the following adaptations were made:

- Dutch related behaviors were implemented in the following subscales:
  1. The item 'makes successful attempts to eat with a knife *and* fork simultaneously' was added (subscale 'use of utensils'- 'functional skills scale').
  2. The items concerning tub transfers with a 'shower' were supplied (subscale 'tub transfers'- 'functional skills scale' and 'caregiver assistance scale').
- Weights and measures were converted to the metric system. For example, the distance in feet (subscale 'outdoor locomotion') was changed into meters; and the size of a half-gallon of milk (subscale 'use of drinking containers') was changed into one liter of milk.
- Literal translated words were changed into Dutch- idiom. For example, the subscale 'bowel management', which was at first translated as 'stoelbeheer', was changed into the less formal word 'poepen'.
- Three new items were added to the following subscales of the 'functional skills scale':
  1. 'Can change position in a bed equipped with a railing' (subscale 'bed mobility/transfers').
  2. 'Walks without support and is able to carry something in the hands at the same time' (subscale 'outdoor locomotion-methods').
  3. 'Devices plans for and plays a game together with another; the playing lasts longer than 30 minutes and is complex' (subscale 'peer interactions').
- Items were more consequently divided into a 'question' and a supportive 'explanatory' part as is shown in table 3.

A specified list of adaptations and motives for these adaptations is presented in the appendix. No items were eliminated from the original PEDI. A bilinguist and American native speaker with experience in translating documents for the biomedical industry then back-translated the Dutch version of the PEDI. She was also previously informed, comparable to the first translator, about the aim of the translation, the criteria and the PEDIs construct.

Table 3. Modification of an item.

	Functional Skills Scale: Problem-Resolution item 23	
	Question	Explanation
PEDI USA version	"When a problem occurs, child can seek help and wait if it is delayed a short time (The child cries "Can't do it!" but calms when told "I can help you in just a minute")"	"The child knowledge that help will be available soon is sufficient to help him/her sustain control when distressed, for short periods of time. However, the child still shows limited tolerance for delays of more than a few minutes."
Dutch PEDI	"If something doesn't work or a problem occurs, the child seeks help and can wait a little while before help is offered."	"For the child, the knowledge that help is coming quickly is enough to be able to manage him/herself for a short time when he/she is upset. However, the child still shows impatience during the few minutes of waiting. For example: The child cries out, "I can't do it!" but calms down when told, "I'm coming to help you."

The Dutch translated version, as well as the back-translated version, a letter of recommendation and a list of adaptations were sent to the authors of the PEDI. They authorized this first draft of the Dutch PEDI.

Content validity

Haley et al.[17] stated that content validity could be considered as an index of whether the items of the PEDI, both individually and as a whole, represent the construct that is supposed to measure. Although a content validity study originally had been performed by Haley et al.[17], because of the adaptations made, the content validity and clinical feasibility of the Dutch translated PEDI had to be re-established.

Methods

Participants

Previously, different allied health professionals familiar with functional status of infants and young children were selected. Thirty-two professionals were solicited to participate, of which 31 (male: female = 4: 27) returned the completed validity questionnaire. None of them was involved in the Dutch adaptation process of the PEDI. One respondent replied anonymously. Professionals represented were pediatric physical therapists (n=15), special educators (n=4), occupational therapists (n=3), physicians at infants healthcare center (n=3), pediatric rehabilitation doctors (n=3), scientific coworkers (n=2), and one speech and language therapist. Eleven respondents were working in a pediatric rehabilitation center, whilst the other respondents were working in a (university-) children's hospital (n=8), a clinic for pediatric physical therapy (n=6), an infants healthcare center (n=3), and at the university (n=3). Mean age of the respondents was 44 years (SD = 9). The professionals average years of experience in child health services was 19 years (SD = 8.82). Fourteen of the respondents had an academic degree in their respective fields (PhD, MA, MSc, MD). Seven of the respondents were familiar with the PEDI.

Validity questionnaire

The validity questionnaire consisted of three sections. In the first section the respondents were asked for their age, highest level of education, working experience, and the degree of familiarity with the PEDI. The second section consisted of eighteen questions. Of those, seven questions could be rated on a nominal scale ('yes/no', 'good/not good'). These questions asked for the accuracy of the content, and the clinical feasibility of the Dutch PEDI. The other eleven questions could be rated on a five-point ordinal scale and asked for the discriminative and evaluative power per domain (ranging from 'very bad' to 'very good'), and the length of the PEDI (ranging from 'too short' to 'too long'). The third section consisted of twelve ques-

tions where respondents could rate the accuracy per item and whether any items should be added or deleted ('yes/no'). Specific feedback information could be written about any of the items.

Analysis

Data are presented for each question by percentages of each response. Written feedback is presented in the results section (qualitative part).

Results

Quantitative

The 18 and 12 questions respectively, of the second and third section of the validity questionnaire, consisted of similar questions for the selfcare domain, mobility domain, and social function domain. For this paper, these questions were clustered into eight topics. These topics are presented in tables 4-11.

Table 4. Topic 1: are the most important facets represented in the particular domains?

	Selfcare	Mobility	Social function
Yes	97%	84%	81%
No	3%	16%	19%

% of respondents that rated this answer.

Table 5. Topic 2: rate the potential of the PEDI to discriminate between non-disabled and disabled children.

	Selfcare	Mobility	Social function	Overall
Very bad	-	-	3%	-
Bad	3%	3%	7%	-
Neutral	10%	7%	29%	13%
Good	68%	84%	58%	84%
Very good	13%	7%	3%	3%
Missing value	7%	-	-	-

% of respondents that rated this answer.

Table 6. Topic 3: rate the potential of the PEDI to identify meaningful change in functional status.

	Selfcare	Mobility	Social function	Overall
Very bad	-	-	3%	-
Bad	16%	13%	26%	10%
Neutral	16%	16%	16%	23%
Good	61%	61%	45%	52%
Very good	7%	7%	3%	7%
Missing value	-	3%	7%	10%

% of respondents that rated this answer.

Table 7. Topic 4: rate the feasibility of the PEDI regarding discriminative and evaluative purposes.

	Discriminative purposes	Evaluative purposes
Not good	3%	10%
Only after modifications	10%	3%
Good	87%	71%
Missing value	-	16%

% of respondents that rated this answer.

For example, 97 % of the respondents found that the most important aspects of the selfcare domain were represented, as to 84% and 81% of the mobility domain and social function domain, respectively (table 4). Eighty-seven percent replied that the potential of the overall PEDI to discriminate in functional status was 'good' (84%) or 'very good' (3%) (table 5). With respect of the evaluative potential of the overall PEDI, a lesser degree of satisfaction (59%) is marked (table 6). For both the discriminative and evaluative potentials, the respondents rated the satisfaction of the social function domain slightly lower than the selfcare and mobility domain. The feasibility of the

Table 8. Topic 5: rate the feasibility to administer the PEDI to parents and professionals.

	Parents/ caregivers	Therapist/ rehabilitation team
Very bad	-	-
Bad	-	16%
Neutral	10%	26%
Good	74%	52%
Very good	13%	7%
Missing value	3%	-

% of respondents that rated this answer.

Table 9. Topic 6: rate the length of the PEDI.

Too short	-
Short	-
Average	13%
Long	71%
Too long	16%

% of respondents that rated this answer.

Table 10. Topic 7: would you add items to the PEDI?

	Selfcare	Mobility	Social function
Yes	32%	42%	23%
No	58%	55%	61%
Missing value	10%	3%	16%

% of respondents that rated this answer.

Table 11 .Topic 8: would you remove items from the PEDI?

	Selfcare	Mobility	Social function
Yes	7%	16%	10%
No	87%	74%	71%
Missing value	7%	10%	19%

% of respondents that rated this answer.

PEDI with respect to discriminative and evaluative purposes was almost rated similarly with the topics before (table 7). Eighty-seven percent rated the feasibility to administer the PEDI on parents/caregivers to be 'good' (74%) or 'very good' (13%)(table 8). This is in contrast with the degree of satisfaction with respect to the feasibility to administer the PEDI on therapists and rehabilitation teams (59%) (table 8). A majority of the respondents is of the opinion that the PEDI is 'long', while 16% rated the PEDI as 'too long' (table 9). Lastly, a majority of the respondents would not add items to the particular domains (table 10), and slightly more respondents would not remove items from the particular domains (table 11).

Qualitative

The majority of the respondents gave specific feedback information, which is presented in this section. With regard to topic 1 (table 4), the most important item that was found to be absent in the mobility domain was 'riding a bicycle'. This was reported by 10 respondents. Other items, less frequently reported, concerned cognitive skills, interactions with the child's siblings, and school functioning. With regard to topic 2 and topic 3 (tables 5 and 6), the respondents reported that the discriminative, and evaluative power might decrease when measuring children with minor functional limitations (such as 'Attention Deficit and Hyperactivity Disorder'), children with a variable degree of disease-severity (as can be found in children with 'Juvenile Idiopathic Arthritis'), children with severe mental retardation, children with autistiforme behavior (such as 'Pervasive Developmental Disorder- Not

Otherwise Specified'), children with sensorial impairments, and very young children as well as children above the target age-group (i.e. between 6 months and 7.5 years). Written comments of some respondents suggested that there are too large steps between the subsequent items to measure clinically meaningful change in functional status. With regard to topic 5 (table 8), respondents reported that the PEDI is probably too detailed to administer it to therapists, in particular because of the lack of information regarding the amount of assistance in home-based activities. General comments were reported concerning the presence of spelling-mistakes and inconsistent usage of language. One respondent missed qualitative aspects of performance, such as starting position and quality of movement.

Discussion

Dutch adaptation of the PEDI

The aim of the adaptation process was to develop a Dutch PEDI, which is the equivalent of the original USA version. Leading guidelines[3,4,6,7] were followed for cross-cultural adaptation. The majority of the adaptations made by the review committee were based on methodological motives, like the re-structuring of the content of the manual in a 'question' and a supportive 'explanatory' part. New items were added because all the members of the review committee agreed that these items would strengthen the construct of the scales. A small number of adaptations were made because of cultural motives, such as the addition of 'the usage of utensils simultaneously' in the subscale 'Use of Utensils', which is suggested to be a culture specific behavior. The fact that a relatively small number of adaptations were made because of cultural motives supported the assumption that the underlying concept of the American PEDI is appropriate to use in the Netherlands.

Content validity

While adaptations were made in the content of the PEDI, it was decided to re-establish the content validity. It also enabled a comparison of our results with the US content validity study, performed in the developing phase of the PEDI[17]. The data of the validity questionnaire indicate that there is at least 81% satisfaction about the overall content of the Dutch PEDI. This is in agreement with the study findings of Haley et al.[17], who reported that 80% of the respondents in the USA were satisfied. Although, the majority of the respondents would not add items in the particular domains, they suggested 'riding a bicycle' frequently as a topic, which could improve the mobility domain.

Bicycling can be considered as an important functional skill in the Netherlands. Brouwers-de Jong et al.[29] reported that a majority of the Dutch children learn to tricycle at pre-school age. Bicycling is the main means of transportation to go to school, friends or shopping centers in the

Netherlands. Therefore, the item bicycling was added to the mobility domain of the functional skills scale. Based on the content validity study, no further items were added to the questionnaire.

A majority of the respondents were of the opinion that the Dutch PEDI has good potentials to discriminate between disabled and non-disabled children. A lower percentage of the respondents, but still a majority, have the same opinion regarding the evaluative potentials, i.e. to measure clinically meaningful change in functional status. In both cases, the social function domain was markedly lower rated than the selfcare and mobility domain. This is in agreement with the study findings of Haley et al.[17] 'Respondents in the US found the PEDI to be a more appropriate index of functional status than a measure of functional change', and 'More concern was expressed about the use of the PEDI as an evaluative instrument in the area of social function (46.4% good or excellent)'[17]. The intention of the authors of the PEDI to include social skills was 'to measure behaviors that had functional relevance, in contrast to developmental tests that are more concerned with achievement of specific components that underlie functional performance'[17]. It is supposed that changes in this more abstracted level of social skills, rendered as behavior, are more difficult to measure over time. Overall, the Dutch PEDI was judged to be a clinically feasible instrument, both for discriminative and evaluative purposes.

The suggestion of the respondents that the discriminative and evaluative potentials are less powerful in specific patient groups, like children with 'Attention Deficit and Hyperactivity Disorder', is in agreement with and supported the directives of the PEDIs manual[10].

It is noteworthy that the PEDI is to be found 'long' or 'too long' by a majority of the respondents. Despite this, the same number of respondents was not prepared to remove any of the items of the particular domains. Some respondents even suggested that there are too large steps between the subsequent items to measure clinically meaningful change in functional status. This paradox was also present in the study of Haley et al.[17]. More (smaller) steps between item levels led to a larger number of items resulting in lengthening the instrument and thus the assessment. This does not serve the practicability. The length of the PEDI might be reduced by selecting a target scale; each PEDI scale is self- containing and can be used separately[10]. Developing instruments is an on-going process. In the future, it will be

necessary to investigate the possibility of whether short forms of the PEDI can be developed for different age classes. This could serve the practicability without loosing the instrument's evaluative power. The opinion of the respondents that the PEDI is more appropriate to be administered to parents as compared to therapists is supported. In our experience using the PEDI, therapists are too less familiar with specific selfcare activities, social function skills and amount of caregiver assistance at home. One respondent missed the point of 'quality of movement' in the judgement whether a child does or does not master the skill. The emphasis of the PEDI however, is to measure the quantitative level of functional status. The suggestions of the respondents regarding spelling-mistakes and inconsistent usage of language were gratefully applied in the revised version. Overall, the expert panel confirmed the functional content of the Dutch PEDI and their comments improved the quality of the content. The corresponding data of the content validity study compared with the results in the USA supported the conceptual equivalence of the Dutch PEDI.

In conclusion, it is stated that the conceptual, semantic, and content equivalence of the Dutch PEDI, as compared with the American version, is established by this cross-cultural adaptation and content validity study. More validity studies (known-group validity, construct validity, responsiveness to change) will be performed to confirm the conceptual equivalence of the Dutch PEDI. As equivalence is also necessary with regard to reliability[6], additional studies (inter-respondent, inter- interviewer, and test-re-test reliability, as well as internal consistency) were started.

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Appendix I Chapter 3

Items added to the Functional Skills scale of the PEDI, based on the adaptation process

- **Question:** "Makes successful attempts to eat with a knife and fork simultaneously".
  Explanation: "Cuts with a knife and fork and brings the food to the mouth".
  (Item 9a, subscale 'Use of utensils', Selfcare domain)

- **Question:** "Can change position in a bed equipped with a railing".
  Explanation: "The child can change the position of his/her head, trunk and limbs".
  (Item 16a, subscale 'Bed transfers', Mobility domain)

- **Question:** "Walks without support and can carry something in the hands at the same time".
  Explanation: "The child walks outside on most sorts of surfaces without needing support from a walking aid or the caregiver and carry a book bag, for example".
  (Item 39a, subscale 'Means of locomotion, outside the house', Mobility domain)

- **Question:** "Devises plans for and plays a game together; the playing lasts longer (30-60 minutes) and is complex".
  Explanation: "The child begins of his own accord a shared activity (such as playing "hide and seek" or marbles) with one or more other children and continues doing exclusively that for a specific time. While playing, the child is able to negotiate with and attune his play to the other child. For example: it is decided who gets to take turn first, who gets assigned a certain roll, etceteras".
  (Item 34a, subscale 'Interactions with other children of the same age', Social Function domain)

Appendix II Chapter 3

Subscale and items added to the Functional Skills scale of the PEDI, based on the content validity study

Subscale 'Bicycling', Mobility domain

Introduction: "These items are related to the child's ability to transport him/herself by means of a bicycle. Leave the aspect of traffic safety conditions out of the evaluation.

- **Question:** "Can ride a (specially adapted) three-wheeler".
  Explanation: "The child is able, without help, to get on and off a (specially adapted) three-wheeler and moved forward by turning the pedals".

- **Question:** "Can ride a bicycle with training-wheels for at least 10m."
  Explanation: "The child is able, without help, to get on and off a bicycle and to ride it independently".

- **Question:** "Can ride a bicycle for at least 50m. ".
  Explanation: "The child is able, without help, to ride 50m. on a bicycle with no training-wheels. Help with getting on and off the bicycle and verbal instructions are permitted".

- **Question:** "Can ride a bicycle without help for at least 100m. ".
  Explanation: "The child is able, without help, to get on and off a bicycle with no training-wheels and to ride it independently".

Reliability of the Dutch 'Pediatric Evaluation of Disability Inventory' (PEDI)

Chapter 4

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Abstract

Objective: Evaluating the reliability of the Dutch version of the 'Pediatric Evaluation of Disability Inventory' (PEDI), an instrument for measuring functional status (capability and performance in self-care, mobility and social function) of young children using parent interviews.

Design: Inter-interviewer reliability was studied after scoring audiotaped interviews by a second researcher. For intra-respondent reliability the same parent was interviewed twice within two weeks; in inter-respondent reliability both parents of a child were interviewed independently within a few days. The proportion of matching scores was computed, and also intra-class correlation coefficients. Cronbach's alpha was calculated on a sample of 63 healthy children aged two.

Subjects: Parents of 64 non-disabled and 31 disabled (various diagnoses) children were interviewed.

Results: The proportion of matching scores varied from .83 to 1 in inter-interviewer reliability, from .43 to 1 in intra-respondent reliability and from .65 to 1 in inter-respondent reliability. ICC's were all but one above .90. Cronbach's alpha was .89 for the self-care domain, .48 for the mobility domain and .87 for the social function domain.

Conclusions: Although some adaptations have to be made, good psychometric properties of the Dutch PEDI are established.

Introduction

The 'Pediatric Evaluation of Disability Inventory' (PEDI) is developed to measure functional status in young children[1]. The PEDI can be used both for discriminative and evaluative purposes, and meets criteria of reliability and validity[1-4]. This instrument, originally developed for the North-American population, is recently translated and cross-culturally adapted for use in the Netherlands[5]. In the adaptation process four new items are added, while other items are adapted, so new reliability studies are designed to establish the psychometric properties of the first draft of the Dutch PEDI. We study inter-interviewer reliability to find out whether items and the accompanying explanation are interpreted in a uniform way. Internal consistency is established to study the extent in which items within a scale are related. Intra-respondent reliability provides an idea of stability of measures. We also study inter-respondent reliability to find out if there are differences in the judgements of both parents, and we focus on the practical implications of these differences.

PEDI

The 'Pediatric Evaluation of Disability Inventory' measures functional status in children aged between a half and seven and a half years. Both the capability of the child (Functional Skills Scale, 201 items) and the amount of help he/she gets from his/her parents (Caregiver Assistance Scale, 20 items) as well as the equipment used (Modifications Scale, 20 items) in daily tasks are measured by a structured interview with parent(s). Functional status is determined in three domains: self-care, mobility and social function. Table 1 gives an overview of topics in the PEDI.

Items in the Functional Skills Scale are dichotomous and are scored either 'capable' or 'not capable'. Summed scores can be computed in every domain and the American version also gives standardized scores. Both the Caregiver Assistance Scale and the Modifications Scale are ordinal scales, ranging from 'totally dependent' to 'totally independent'.

Table 1. Content of the first draft of the Dutch PEDI

Functional Skills Scale		Caregiver Assistance Scale
Subscales	**Number of items**	**Modifications Scale**
Self care		
Types of food textures	4	Eating
Use of utensils	5 + 1#	
Use of drinking containers	5	
Tooth brushing	5	Grooming
Hair brushing	4	
Nose care	5	
Hand washing	5	Bathing
Washing body and face	5	
Pullover/front-opening garments	5	Dressing upper body
Fasteners	5	
Pants	5	Dressing lower body
Shoes/socks	5	
Toileting task	5	Toileting
Management of bladder	5	Bladder Management
Management of bowel	5	Bowel Management
Mobility		
Toilet transfers	5	Chair/toilet transfers
Chair/Wheelchair transfers	5	
Car transfers	5	Car transfers
Bed mobility/transfers	4 + 1#	Bed mobility/transfers
Tub transfers	5	Tub transfers
Indoor locomotion methods	3	Indoor locomotion
Indoor locomotion – distance/speed	5	
Indoor locomotion – pulls/carries objects	5	
Outdoor locomotion methods	2 + 1#	Outdoor Locomotion
Outdoor locomotion – distance/speed	5	
Outdoor surfaces	5	
Up stairs	5	Stairs
Down stairs	5	

Table 1 continued

Functional Skills Scale		Caregiver Assistance Scale
Subscales	**Number of items**	**Modifications Scale**
Social function		
Comprehension of word meanings	5	Functional
Comprehension of sentence complexity	5	comprehension
Functional use of communication	5	Functional expression
Complexity of expressive communication	5	
Problem – resolution	5	Joint problem-solving
Social interactive play (adults)	5	
Peer interactions (child of similar age)	5 + 1[#]	Peer play
Play with objects	5	
Self information	5	
Time orientation	5	
Household chorus	5	
Self protection	5	Safety
Community function	5	

[#]: items added in the Dutch version

Methods

Participants

In pre-testing the Dutch PEDI we interviewed parents of children with disabilities ($n = 31$) and without known disabilities ($n = 64$). Children without known disabilities were between two and three years old. A first group consisted of children visiting a primary healthcare center for infants where growth and development of healthy children is watched routinely. Parents of all children aged two, living in a small town in the center of the Netherlands ($n = 260$) received a letter from the healthcare center with an outline of the study and a request to participate. Parents of 44 children were interviewed (a response of 17%). From nine children both parents were interviewed and 11 interviews were audiotaped so that a second researcher also could score the interview. A second group of non-disabled children was already participating in another study when they were asked to participate in this study: 20 of them agreed.

In this study, children with disabilities are known to a specific Children's hospital. They are having neurometabolic conditions ($n = 13$), Spina Bifida ($n = 7$), Osteogenesis Imperfecta ($n = 6$) and Infantile Encephalopathy ($n = 5$). All children have stable or slowly progressive limitations in performing daily activities.

We audio-taped 21 interviews with parents of children with disabilities for the inter-interviewer reliability, from 18 children both parents were interviewed and 20 parents were interviewed twice to establish intra-respondent reliability.

Analysis

Inter-respondent and inter-interviewer reliability was established using scores from both disabled and non-disabled children. Excluded from the reliability analysis were those interviews with summated skill scores on a particular domain less than 10% or more than 90% of the maximum summated score on that domain. Consequently, we excluded nine interviews in the

selfcare domain, 39 interviews in the mobility domain and eight interviews in the social function domain. The exclusion of the mobility domain in 39 interviews was due to the nearly maximum scores of non-disabled children. In the other domains exclusion was due to very low scores of disabled children.

In all reliability studies we first looked at the proportion of same answers in every item. Although Cohen's kappa is the usual measure of correspondence, its size depends on variance in answers. However, dichotomous skill-questions didn't always vary (a lot) because of a rather homogenous group of non-disabled children. Therefore, we considered the proportion of same answers in every item instead of Cohen's kappa. In addition intra-class correlation coefficients (ICC) are calculated.

In studying intra respondent reliability the same parent of the same child was (partially) interviewed twice: mean time between the two interviews was 14.9 days (sd 3.6 days). In studying inter-respondent reliability parents were requested not to inform each other about the interview before both interviews were finished. The mean time between the interviews was 3.9 days (sd 4.8 days).

In table 2 the number of used interviews in every domain is shown.

Table 2. Number of used interviews

	Interinterviewer		Intra respondent		Inter-respondent		Internal consistency	
	F	C/M	F	C/M	F	C/M	F	C/M
Self care domain	24	23	7	6	26	26	63	63
Mobility domain	23	21	9	8	20	20	34	34
Social function domain	25	23	10	10	27	27	63	63

Functional Skills Scale (F), Caregiver Assistance Scale (C) and Modifications (M) scale

Results

A) Functional skills scale

Inter interviewer reliability

The proportion matching scores varied from .83 to 1. No differences were found in 44 of the 74 items of the self-care domain (59%), in 49 of the 61 items of the mobility domain (80%) and in 32 of the 66 items of the social function domain (48%). Intra-class correlation coefficients were .99 for all domains. As all interviews were audiotaped, it was possible to determine the 'right' score on items where differences exist, the score in according with the answer of the parent. In one third of the differences, especially in items were correspondence was relatively low, differences resulted from ambiguous interpretation of the item and/or the explanation. These items or explanations will be adapted in the final version. For example, item 41 in the social function domains is 'Can say its own name'. Is a child capable only when he/she pronounces his/her name correctly, or also when he/she calls him/herself consequently in the same manner but not correct (which is often the case when a child has a name that is hard to pronounce)? In the other two third of the differences, one of the researchers scored inaccurate. Sometimes it was very obvious, in other cases parents gave an explanation after their judgement 'capable' or 'not capable' by which the initial answer turned out to be incorrect in terms of the scoring criteria. Both the researcher that took the interviews and the researcher who scored the audiotaped interviews sometimes had inaccurate scores, and no systematic differences were found between interviewers in scoring 'capable' or 'not capable'.

Intra respondent reliability

The proportion matching scores varied from .43 to 1. No differences were found in 45 of the 74 items of the self-care domain (61%), in 57 of the 61 items of the mobility domain (93%) and in 52 of the 66 items of the social

function domain (79%). Intra class correlation coefficients were .98 for all domains. The mean differences between first and second interview were for the self-care domain 1 (sd 3.3), for the mobility domain .67 (sd 2.7,) and for the social function domain -2.2 (sd 3.8). Those parents who wanted to re-do the whole interview had the most extreme difference scores.

Inter respondent reliability

The proportion matching scores varied from .65 to 1. No differences were found in 11 of the 74 items of the self-care domain (15%), in 15 of the 61 items of the mobility domain (25%) and in 8 of the 66 items of the social function domain (12%). Intra class correlation coefficients were .95 for the self-care and mobility domain, and .91 for the social function domain. In case of differences mothers more often (about 60%) scored 'capable' than fathers did, in all domains. Analyzing data of disabled and non-disabled children separately showed that for non-disabled children mothers and fathers are equally positive, while for the disabled children mothers scored 'capable' twice as much as fathers did.

Internal consistency

Cronbach's alpha was computed for the sample of 63 non-disabled children, both for the three domains and for sub-scales within these domains. For the self-care domain, $\alpha = .89$, for the mobility domain $\alpha = .48$ and for the social function domain, $\alpha = .87$. Cronbach's alpha for the sub-scales varied from 0 to .87.

Table 3. Summed scores: mean, standard deviation and T-test

		Inter interviewer		Intra respondent		Inter respondent	
		Researcher 1	Researcher 2	First interview	Second interview	Mother	Father
Self-care	Mean	40.1	39.7	27.3	26.3	37.6	36.0
	Sd	16.6	16.2	16.7	16.7	14.5	15.6
	t-test	t = .81 (P = .429)		t = .80 (P = .455)		t = 1.83 (P = .079)	
Mobility	Mean	43.5	43.4	33.8	33.1	32.5	31.5
	Sd	11.1	11.2	16.6	17.1	15.8	14.9
	t-test	t = .53 (P = .606)		t = .73 (P = .486)		t = .89 (P = .386)	
Social function	Mean	33.8	33.6	33.4	35.6	37.7	35.8
	Sd	17.4	17.0	16.2	13.8	13.0	14.1
	t-test	t = .47 (P = .646)		t = -1.83 (P = .100)		t = 1.72 (P = .097)	

B) Caregiver Assistance Scale

Inter interviewer reliability

In all but 2 items differences were found between scores of both researchers. The size of the differences was minimal most of the time (61 of the 74 differences). In the self-care and social function domain researcher 1 scored higher (i.e. more independent) than researcher 2; for the mobility domain, on the contrary, researcher 2 scored more independent. Intra-class correlation coefficients were .96 for the self-care domain, .98 for the mobility and the social function domain.
Despite the fact that the interviews were audiotaped, it was not always possible to determine the score that best fit the parent's answer. The supportive explanatory part of the item was not always clear specifically in items concerning different activities where it was not clear how to relate those activities into one score. These items will be adapted.

Intra respondent reliability

In all but three (self-care) items differences were found between scores from the first and second interview. The size of the differences was minimal most of the time (38 of the 48 differences). In the self-care domain most differences show progression in Caregiver Assistance (lower scores, children were judged less independent), while in the social function domain most differences showed a decline in Caregiver Assistance and in the mobility domain both progression and decline were found. Intra-class correlation coefficients were .97, .94 and .91 for the self-care, mobility and social function domain respectively.

Inter respondent reliability

In all items differences in scores of father and mother were found. The size of the differences was minimal most of the time (138 of the 186 differences). In all domains, mothers judged their child as more independent than fathers

did. Intra-class correlation coefficients were .94, .93 and .79 for the self-care, mobility and social function domain respectively.

C) Modifications Scale

Inter interviewer reliability

Most of the differences in the Modifications Scale were in the self-care domain, especially in items G and H (bladder and bowel management) where researcher 1 often forgot to score diapers. In the other items researcher 1 and 2 varied in scoring the equipment used regularly. Sometimes it was clear, in other cases it is hard to determine how to score the equipment used. For example an electric toothbrush is not a modification as meant, but it is when a parent chooses to use an electric toothbrush for his/her child because of its functional limitations. Almost no differences were found in the social function domain because modifications in communication (asked for in four of the five items) are only used when children are hearing or speech disabled, and our sample didn't contain such children.

Intra respondent reliability

Few differences concerning modifications were found between the first and second interview. The small differences found were nearly all changes from 'no modifications' in 'child-oriented modifications'.

Inter respondent reliability

Differences were found both in the self-care and in the mobility domain; fathers and mothers varied in their judgement of equipment used. Sometimes the father mentioned extensive modifications and the mother no modifications, and vice versa. In almost half of the differences father and mother had very different ideas about the equipment used.

Discussion

The purpose of this study was to examine the reliability of the first draft of the Dutch PEDI. Intra-class correlation coefficients were high, so reliability was established, and by considering not only scales but also individual items we could improve some of these items in the final version.

Haley[1] described the psychometric properties of the original PEDI. Cronbach's alpha was computed for the normative sample (n = 410): for the Functional Skills Scale alpha was .99, .97 and .98 for the self-care, mobility and social function domain respectively. We decided to control for maturation by computing alpha on a sample that was homogeneous for age, and we found lower alphas (.89, .48 and .87 respectively). The explanation for the low alpha in the mobility domain is the exclusion of 39 interviews because of very high-summated scores: when Cronbachs alpha was computed with all the 63 subjects the value was .74. Dutch research with children of other ages must confirm the high alpha's of the original PEDI.

In all parts of this study, except for the internal consistency, the mobility domain scored better than the self-care and social function domain. An explanation might be that items of the mobility domain, more than in other domains, are less subjected to choices parents make when raising their children. For example, carrying small objects (mobility, item 35) is different from 'using a knife' (self-care, item 9) where the choice of parents whether or not to let their child practice with a knife is, in some age-groups, the main explanation for the score 'capable' or 'not capable'.

Why did interviewers not always score in agreement with the parent's answers? Both researchers did score inaccurately, so we do not think it is due to an interviewer effect. Test-length might be a factor that can partially answer the question. When interviewing a parent, for every topic in the Functional Skills Scale, the researcher makes a choice what item is offered first to the parent, based on the age and disability of the child. The score on that item determines what the next item will be: if the child scores 'capable' on that first item, the interviewer offers a more difficult item, if the score is 'not capable', the interviewer offers a less difficult item. Despite the fact that in this way not all 201 Functional Skills Scale items are offered, the PEDI-interview lasts about 45 to 60 minutes and it seems hard to be totally

concentrated all that time. Because parents not always answer in accordance with the scoring criteria, more information than a simple 'capable' or 'not capable' is required to be sure that the parent has understood the question well and the child meets the scoring criteria. Respondents in the content validity study[5] also mentioned the large length of the test as a possible disadvantage of the PEDI, but did not want to remove items.

In the inter-respondent study it became clear that parents differ in their judgement of the functional status of their child. Mothers judged their children more capable and less dependent on their assistance than fathers did, especially when the child is disabled. Is it because mothers spent more time taking care of their children than fathers did? Both variables, sex and the extent to which a parent takes care of the child (comparing with the other parent), were highly related. We can't conclude that the judgement of the mother is better than the judgement of the father, because the mother spends more time with her child. But when the PEDI is used for evaluative purposes, it's important to interview the same parent, to avoid that differences between two measures are the result of differences in judgement between parents rather than changes in functional status over time.

Because reliability was confirmed in this study, the Dutch PEDI can be used in further research to establish validity and in making standardized scores for the Dutch population.

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Discriminative validity of the Dutch 'Pediatric Evaluation of Disability Inventory' (PEDI)

Chapter 5

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Abstract

Objective: To examine the discriminative validity of the Dutch 'Pediatric Evaluation of Disability Inventory' (PEDI) to discriminate functional status between children with and without disabilities.

Design: Cross-sectional study.

Setting: University Children's Hospital

Patients and participants: A clinical sample comprising 197 children with disabilities (Infantile Encephalopathy, N= 40; Juvenile Idiopathic Arthritis, N= 20; Neurometabolic conditions, N=36; Neuromuscular Disorders, N= 9; Skeletal Disorders, N= 28; Spina Bifida, N= 41; Traumatic Injury, N= 23), and 62 children without disabilities participated in this study.

Outcome Measure: Functional status was measured using a Dutch adapted version of the 'Pediatric Evaluation of Disability Inventory' (PEDI).

Results: Discriminant analysis established the sensitivity and specificity of the PEDI. Correct predictions of group-membership (disabled vs. non-disabled) were found in both children without disabilities (93% correctly predicted) and children with disabling conditions (91% correctly predicted).

Conclusion: The discriminative validity of the Dutch PEDI between children with and without disabilities was found to be excellent.

Introduction

The 'Pediatric Evaluation of Disability Inventory' (PEDI), developed by Haley et al.[1,2] is a clinical instrument for the assessment of functional status in children from 0.5 to 7.5 years of age. It is a judgement based structured interview for parents used by professionals in rehabilitation medicine and in health related outcome research. The PEDI is able to measure both *capability* (what the child can do) and *performance* (what the child actually does do) of routine daily childhood activities. It is comprised of three content domains: selfcare, mobility, and social functioning, resulting in 6 outcome scales.

A main goal of the PEDI is to detect whether a functional deficit or delay exists in children with respect to the functional status development, and if so, the extend and content area of the delay or deficit. The PEDI can be viewed as a discriminative outcome instrument according to the classification of Kirshner and Guyatt[3]. Feldman et al.[4] examined the construct validity with respect to the discriminative power of the PEDI. They compared the outcome of the PEDI between 20 children with disabilities, i.e. children with arthritic conditions and spina bifida, and a matched normative sample. The children with disabilities scored significantly lower than the children without disabilities in the selfcare- and mobility domain. Although the results confirm the potentials of the PEDI to discriminate, it was based on a small sample size. Other kinds of validity studies and reliability studies were published[2,5-8] with the original PEDI.

The applicability of the PEDI for Dutch society was examined in a preliminary study[9]. PEDI scores of Dutch children without disabilities were compared with American peers. The results showed different profiles in outcome indicating possible inter-cultural differences. A Dutch translation and adaptation of the PEDI[10] was subsequently conducted based on current scientific guidelines in cross-cultural research[11-13]. Four new items were added to the original PEDI while some of the 197 existing items were adapted or re-formulated in order to fit better for the Dutch society. Examples of these adaptations are the conversion of weights and measures into the metric system, and the addition of a 'shower' to the items concerning 'tub transfers'. No items were eliminated from the original PEDI. The authors of the

original PEDI authorized the content of the Dutch PEDI. At the moment, the Dutch PEDI still has to be calibrated for Dutch children in the age group 0.5-7.5 years.

In this study we examined the discriminant validity of the Dutch PEDI to complete the adaptation process. Children with and without disabilities were included, and discriminant analysis was used to examine whether the Dutch PEDI was able to correctly identify children with functional deficits. The choice of the clinical sample was based on the assumption that a broad spectrum of functional limitations, physically and/or intellectually, was needed to capture the whole PEDI content. Therefore, we included children with central-nervous system (CNS) impairments and children with musculo-skeletal impairments.

Regarding the first group we included children with a known psychomotor delay, Spina Bifida and Infantile Encephalopathy. It was assumed that functional limitations would be found in physical as well as cognitive domains of the PEDI in the children with CNS involvement since intellectual impairments are not uncommon in these patient groups. In addition, children with Juvenile Idiopathic Arthritis, Osteogenesis Imperfecta, traumatic injury, and neuromuscular disorders represented children with musculo-skeletal involvement. In these children, it was assumed that functional limitations would be found mainly in ambulation and selfcare skills. Although it was not the main purpose of the study, we looked as well at differences between the clinical groups as a sideline.

Methods

Subjects

Between August 1999 - November 2000, 62 children without disabilities were recruited from a health-care center for infants and toddlers (Table 1). Parents visited this outpatient clinic for routine health assessment of their child. A clinical sample was measured between January 1999 - October 2000 comprising 197 children with different kinds of disabilities (Table 1). Of them, 166 children were recruited from the University Children's Hospital and from an affiliated rehabilitation center, while the other 31 children were recruited from a study in children with Infantile Encephalopathy. All children were approached after a (first) visit to the outpatient's clinic within the given time frame of the study.

Table 1. Patient characteristics.

Clinical group	Age Mean (sd)	Age Range	Boys	Girls	Total
NeuMet	35.2 (17.8)	10-87	22	14	36
SpiBif	42.9 (25.6)	10-89	18	23	41
SkelDis	58.1 (23.5)	23-90	13	15	28
InfEnc	64.7 (17.2)	23-90	28	12	40
JIArthr	39.0 (21.5)	14-88	6	14	20
Trauma	44.0 (21.1)	10-84	13	10	23
NeuMus	70.4 (12.5)	49-84	5	4	9
All Dis	49.0 (23.8)	10-90	105	92	197
Non Dis	30.6 (3.8)	24-35	26	36	62
Total			131	128	N=259

Age in months; NeuMet (Neurometabolic conditions), SpiBif (Spina Bifida), SkelDis (Skeletal Disorders), InfEnc (Infantile Encephalopathy), JIArthr (Juvenile Idiopathic Arthritis), NeuMus (Neuromuscular Disorders), All Dis (All children with a disability), Non Dis (Children without a disability).

The clinical sample comprised seven diagnostic groups (Table 2). The children were previously diagnosed, with the exception of the children with symptoms of a neurometabolic disorder. These children, in which there was not always a diagnosis at hand, presented different levels of psychomotor delay, sometimes associated with seizures, muscular conditions, failure to thrive, and sensory impairments.
Children and parents were excluded if they were not able to actively use the Dutch language. This was determined at the introduction of the study when they were not able to iterate what they were told about the procedure.

Instrument

The child's functional capability was measured using 3 *functional skills scales* of the Dutch PEDI[2,4]. These scales contain a total of 201 questions organized within 41 subscales concerning 3 domains: selfcare domain, mobility domain, and social function domain (Table 3a). Each question is scored positive (score 1) or negative (score 0). A positive score was given when a child had mastered the particular skill. Raw scores for each subscale and per domain were summed.
The child's performance was measured using 3 *caregiver assistance scales* of the Dutch PEDI[2,4]. These scales contain 20 questions concerning the same activities of the functional skills scales (Table 3b). The amount of assistance is scored on a 6-point ordinal scale. Scores of 0 and 1 refer to the supportive participation of the caregiver for more than half of the activities, while scores of 2 to 5 refer to a progressive independence of the child. Raw scores were summed for each domain.

Procedure

Five experienced clinicians, chosen for their expertise on the relevant patient groups interviewed the parents. The interviewers completed a training program according to the guidelines of the PEDI manual[2]. All parents (N= 259) who participated in the study gave informed consent. The Dutch PEDI was administered at home or in the hospital in 105 cases, 154 inter-

Table 2. Clinical samples characteristics.

Clinical group	Subtype			
Spina Bifida * (n= 41)	Thoracic lesion (n=7)	Lumbar 1-3 lesion (n=8)	Lumbar 4-5 lesion (n=15)	Sacral 1-2 lesion (n=11)
Skeletal Disorders (n= 28)	Osteogenesis Imperfecta type 1 (n= 17)	Osteogenesis Imperfecta type 3 (n= 5)	Osteogenesis Imperfecta type 4 (n= 4)	Achondrodysplasia (n= 2)
Infantile Encephalopathy (n= 40)	Hemiplegia (n= 21)	Diplegia (n= 14)	Quadriplegia (n= 4)	Others (n= 1)
Juvenile Idiopathic Arthritis (n= 20)	Mono-articular (n= 2)	Oligo-articular (n= 11)	Poly-articular (n= 4)	Systemic (n= 3)
Trauma (n= 23)	Upper extremity (n= 9)	Lower extremity (n= 12)	NeuroTrauma (CNS) (n=2)	
Neuromuscular Disorders (n=9)	Anterior horncell (n= 1)	Peripheral nerve (n= 3)	Muscular (n= 5)	

* Spina Bifida with myelomeningocèle (n=41) and shunted hydrocephalus (n = 39).

Table 3a. Item content of the Dutch PEDI.

Self-Care Domain *	Mobility Domain †	Social Function Domain ‡
Types of food textures	Toilet transfers	Comprehension of word meanings
Use of utensils	Chair/wheelchair transfers	Comprehension of sentence complexity
Use of drinking containers	Car transfers	Functional use of communication
Tooth brushing	Bed mobility/transfers	Complexity of expressive communication
Hairbrushing	Tub transfers	Problem-resolution
Nose Care	Indoor locomotion- methods	Social interactive play
Hand washing	Distance/speed indoors	Peer interactions
Washing body and face	Pulls/carriers objects	Play with objects
Pullover/front-opening garments	Outdoor locomotion- methods	Self information
Fasteners	Distance/speed outdoors	Time orientation
Pants	Outdoor surfaces	Household chores
Shoes/socks	Upstairs	Self protection
Toileting tasks	Downstairs	Community function
Management of bladder		
Management of bowel		

Functional skills scale; * Selfcare domain: 74 questions in 15 subscales; † Mobility domain: 61 questions in 13 subscales; ‡ Social Function domain: 66 questions in 13 subscales.

Table 3b. Item content of the Dutch PEDI.

Self-Care Domain *	Mobility Domain †	Social Function Domain ‡
Eating	Chair/ toilet transfers	Functional comprehension
Grooming	Car transfers	Functional expression
Bathing	Bed mobility/transfers	Joint problem solving
Dressing upper body	Tub transfers	Peer play
Dressing lower body	Indoor locomotion	Safety
Toileting	Outdoor locomotion	
Bladder management	Stairs	
Bowel management		

Caregiver assistance scale; * Selfcare domain: 8 subscales; † Mobility domain: 7 subscales; ‡ Social Function domain: 5 subscales.

views were administered by telephone. To improve validity, we administered the PEDI to the most proxy caregiver. This was left to the judgement of the caregivers and was subsequently reported.

The parents of children with Juvenile Idiopathic Arthritis were interviewed within a month after they visited the outpatients department for the first time. Because symptoms may vary from day to day in children with Juvenile Idiopathic Arthritis, we standardized the interview by asking the parents to base their judgement on the past 14 days. Parents of children with a traumatic injury were interviewed within 14 days after the incident, and they were asked to base their judgement on the actual functional status. All other interviews were done during their visit to the outpatient's department or within one month.

Data Analysis

Based upon an analysis of covariance[14] age corrected scale scores were computed for each of the 6 outcome scales (3 functional skills scales and 3 caregiver-assistance scales). This was necessary in order to correct for age differences among the eight groups (Table 1).

Discriminant validity has been examined using discriminant analysis, after we established the reliability and item-test correlations of each of the 6 outcome scales (which were around .94 and .78, respectively). Discriminant analysis was conducted by canonical discriminant functions[14], and was used to predict a child's group membership using his or her 6 age-corrected scales scores. The SPSS statistical program, version 7.5 was used for the analysis.

Results

Table 4 presents the resulting cross-tabulation of observed and predicted group membership. The diagonal (bold) is representing the amount of correctly identified children in their respective (clinical) groups, based on the PEDI outcome. We found that 93.5 % of the children without a disability were correctly predicted as being non-disabled, and 8.4 % of the children with a disability were predicted as *not* having a disability, i.e. 91.6 % of the children with a disability were correctly predicted as being disabled.
Regarding the clinical samples, the best predictions were made in children with neurometabolic conditions. Our data also seems to indicate that our predictions cluster into two major groups: CNS involvement or musculoskeletal involvement.

Table 4. Classification results.

	Predicted Group Membership								
Observed	NonDis	NeuMet	SpiBif	SkelDis	InfEnc	JIArthr	Trauma	NeuMus	Total
NonDis	**93.5**	1.6	.0	1.6	.0	3.2	.0	.0	100.0
NeuMet	5.6	**75.0**	.0	.0	8.3	8.3	.0	2.8	100.0
SpiBif	4.9	24.4	**39.0**	7.3	7.3	2.4	7.3	7.3	100.0
SkelDis	10.7	3.6	7.1	**21.4**	17.9	21.4	14.3	3.6	100.0
InfEnc	10.0	22.5	10.0	5.0	**42.5**	2.5	5.0	2.5	100.0
JIArthr	10.0	5.0	10.0	.0	.0	**50.0**	5.0	20.0	100.0
Trauma	17.4	8.7	4.3	4.3	8.7	4.3	**43.5**	8.7	100.0
NeuMus	.0	.0	.0	.0	11.1	22.2	22.2	**44.4**	100.0
AllDis	**8.4**								

Predicted group membership in percentiles. NonDis (Children without a disability), NeuMet (Neurometabolic conditions), SpiBif (Spina Bifida), SkelDis (Skeletal Disorders), InfEnc (Infantile Encephalopathy), JIArthr (Juvenile Idiopathic Arthritis), NeuMus (Neuromuscular Disorders), AllDis (All children with a disability).

Discussion

The aim of discriminative measures is to distinguish between individuals or groups on underlying dimensions[3]. Discriminative measures in rehabilitation medicine are useful to determine the impact of a disorder with respect to functional status at a single point of time.

The purpose of this study was to examine the discriminative validity of the Dutch adapted PEDI, i.e. the ability of the Dutch PEDI to discriminate between children with and without disabilities with respect to functional status. This question was solved using discriminant analysis. However, we first performed analysis of covariance and computed age-corrected scale scores, because discriminant analysis could not be conducted in this study with children of the same age because of too small sample sizes.

As 93.5% of the children without disabilities were correctly predicted as being non-disabled (based on the PEDI outcome), and 91.6% of the children with disabilities were correctly predicted as being disabled, we conclude that the Dutch PEDI discriminates excellent between children with and without disabilities. It confirms a high degree of *sensitivity* (correct identification of children with disabilities within this population) and *specificity* (false prediction of children without disabilities who were identified as disabled).

In our study, we were not able to compare the differences between patient groups extensively due to inhomogeneous samples regarding sample size, varying degrees of disease severity within a group, and lack of data with respect to intellectual skills of the children. However, when clustering diagnostic groups with a known CNS involvement, i.e. psychomotor delay, Spina Bifida, and Infantile Encephalopathy, and diagnostic groups with a known musculo-skeletal involvement, i.e. Juvenile Idiopathic Arthritis, Osteogenesis Imperfecta, traumatic injury, and neuromuscular disorders, discriminant validity shows equally high prediction rates, i.e. 76.0% and 67.5% (Table 5). This is at least suggestive for further support of the good discriminant validity of the Dutch PEDI.

When calculating standardized means of all six subscales in all (diagnostic) groups, it becomes evident that the functional status of the subset of patients with a hypothesized musculo-skeletal involvement, appears to be

Table 5. Classification results.

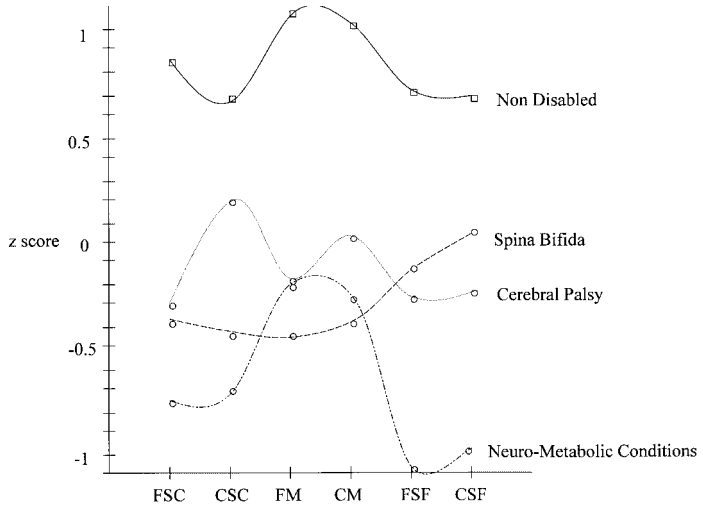
	Predicted Group Membership			
Observed	NonDis	CNS	MS	Total
NonDis	**93.5**	1.6	4.9	100.0
CNS	6.8	**76.0**	17.2	100.0
MS	11.3	21.2	**67.5**	100.0

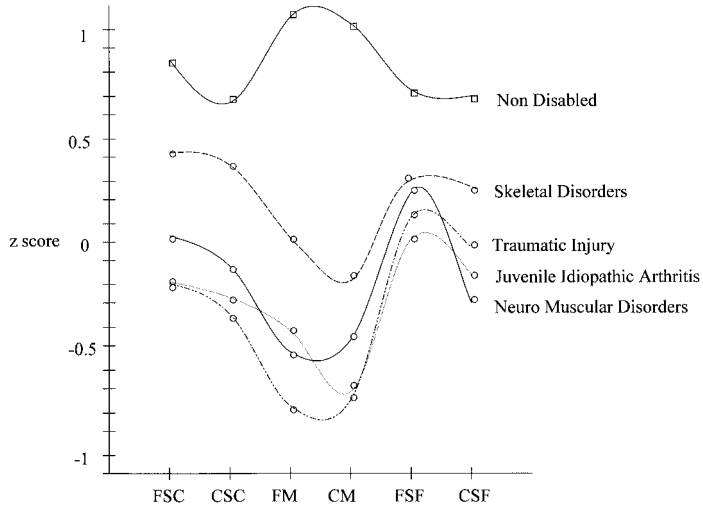
Predicted group membership in percentiles. NonDis (Children without a disability), CNS (Central Nervous System involvement: Psychomotor delay, Spina Bifida, Infantile Encephalopathy), MS (Musculo-skeletal involvement: Skeletal Disorders, Juvenile Idiopathic Arthritis, Traumatic injury, and Neuromuscular Disorders).

more homogeneous compared to the subset of patients with a hypothesized CNS involvement (Figures 1a and 1b). Especially on PEDI subscales susceptible to cognitive function. This finding underlines the need for further studies in homogeneous diagnostic groups to analyze the PEDI's discriminative validity between different groups. Figure 1a and 1b enable us also to visualize the discriminative validity between children with and without disabilities.

In conclusion, this study confirmed the discriminative validity of the Dutch PEDI. It discriminates excellent between children with and without disabilities with respect to functional status of daily activities. The results established the applicability of the PEDI for discriminative purposes in the patient groups, which were used in this study. Therefore, the PEDI can serve as a diagnostic tool for professionals in pediatric rehabilitation medicine.

Figure 1a. + 1b. Standardized scores for age-corrected scales.





Vertical axis: Z-scores; horizontal axis: 6 outcome scales of the PEDI.

FSC (Functional Skills Scale- Selfcare domain), CSC (Caregiver Assistance Scale- Selfcare domain), FM (Functional Skills Scale- Mobility domain), CM (Caregiver Assistance Scale- Mobility domain), FSF (Functional Skills Scale- Social function domain), CSF (Caregiver Assistance Scale- Social function domain).

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Responsiveness of the 'Dutch Pediatric Evaluation of Disability Inventory' (PEDI): A new approach in responsiveness studies

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Chapter 6

Abstract

A well-accepted method in examining the responsiveness of a new instrument is a multiple-group design, in which changes in outcome over time will be measured at group level. An increased interest in patient-centered health services started the discussion how to examine the responsiveness of an instrument at individual level. The purpose of this study was to examine the responsiveness of the 'Pediatric Evaluation of Disability Inventory' (PEDI), a functional status instrument for infants and young children. Children with Traumatic Injury (TI), Neuromuscular Diseases (NMD), and Juvenile Idiopathic Arthritis (JIA) (N=85) were measured three times. A Global Rating Scale (GRS) was used as an external standard of change, and analyses were done at an individual level. Internal and external responsiveness were tested using 'order restricted statistical inference'. The internal responsiveness of the PEDI varied from moderate to excellent. The external responsiveness varied from moderate to good. The agreement between GRS and PEDI differed for each child, while responsiveness was dependent of subjects, clinical sample, and the choice of the external standard. This study further widens the approach in measuring the responsiveness of instruments at an individual level.

Introduction

Evaluative instruments in health outcome studies are designed to measure changes within patients over time[1]. These instruments should be able to measure improvement or deterioration in health status. Another essential characteristic of an evaluative instrument is that changes in scores should not occur when the subject's health status is stable (i.e. a low intra-subject variation or high level of reliability). Changes in scores should reflect true changes in health status rather than changes as the result of measurement errors[1-4].

In developing an evaluative instrument it is common to investigate responsiveness. Strategies for assessing an instrument's responsiveness are reviewed in current literature[2,5-7]. Stratford et al.[2] prefer to use multiple-group designs in which two or more patient groups have to be compared, and health status is expected to change to varying degrees. The most favorable design is to compare the outcome of two randomly selected patient groups, in which one will receive a placebo treatment, whilst the other will receive a previously proven intervention[2,8]. This design, however, requires a homogeneous sample of subjects.

There is no general agreement regarding the statistical approaches in examining the responsiveness. Wright and Young[6] tested several responsive outcome instruments using five widely known statistical indices. Each index reflected another rank ordering of the most responsive instrument. In contrast to statistical approaches, there is consensus regarding the need of a 'golden', or external standard, which supports the changes that are found. External standards for example, should contain global ratings of patients' satisfaction and/or disease severity, laboratory tests, or other instruments measuring similar concepts.

More recently, Husted et al.[7] introduced the distinction between 'internal responsiveness' and 'external responsiveness'. Internal responsiveness refers to the ability of an instrument to detect changes, while external responsiveness reflects to 'the extent to which changes in a measure relate to corresponding changes in a reference measure'.

In the last decade, it became more and more important to aim at patient-centered health services. One might expect that validity studies with

respect to responsiveness, or intervention studies were aimed at changes in health status at the individual level. Surprisingly, most of these studies were concerned with the evaluative potentials of the instrument or the effects of interventions in patient groups[2,6,9-15].

In this study, a multi-group design was applied to measure the responsiveness of the Dutch version of the 'Pediatric Evaluation of Disability Inventory' (PEDI)[16,17]. The outcome of two patient groups was measured with known, but different natural courses in functional status development. A global rating scale (GRS) was used as an external standard to support the expected different directions (improvement or deterioration), and the amount of changes that would occur in the particular groups. However, the PEDI results and the GRS were analyzed at an individual level. This paper widens the approach in measuring the responsiveness of instruments in patient-centered health outcome.

PEDI

The 'Pediatric Evaluation of Disability Inventory' (PEDI), developed by Haley et al.[16,18] is a clinical instrument for the assessment of functional status in children up to 7.5 years of age. It is a judgement based structured interview, which can be administered to parents. The PEDI is able to measure both *capability* (what the child can do) and *performance* (what the child actually does do) of routine daily childhood activities. It aims at three content domains: selfcare, mobility, and social functioning, resulting in 6 outcome scales. The PEDI is primarily designed for functional evaluation of young children, but can be used for the evaluation of older children if their functional abilities fall below that of children up through the age of 7.5 years. We refer to published materials for an extensive description of the content and psychometric properties of the PEDI[16,19-24].

Methods

Subjects

A clinical sample comprising 85 children was measured between April 1999 - April 2001. They were recruited from the University Children's Hospital and an affiliated pediatric rehabilitation center. The children were diagnosed with Juvenile Idiopathic Arthritis (JIA, N=20), Traumatic Injury (TI, N=44), or Neuro-Muscular Disease (NMD, N= 21) (Table 1).

Instrument

The Dutch version of the PEDI[17] was used to measure the functional status of the children. The capability of the children was measured using *three functional skills scales*[16]. These scales contain a total of 201 questions, organized within 41 subscales, concerning three domains of daily activities: selfcare, mobility, and social functioning. Each question had to be scored positive (score 1) or negative (score 0). A positive score was given when a child had mastered the particular skill. Raw scores per subscale were summed.
Performance of the child was measured using three *caregiver assistance scales*[16]. These scales contain 20 questions concerning the same activities as the functional skills scales. The amount of assistance was scored on a 6-point ordinal scale. Scores 0 and 1 referred to the supportive participation of the caregiver for more than half of the activities, while scores 2 to 5 referred to a progressive independence of the child.
After data collection, we clustered the subscales from the six outcome scales into nine main topics: Eating/Drinking, Grooming/Bathing, Dressing, Toileting, Indoor locomotion and Transfers, Outdoor locomotion and Transfers, Communication, Play and Interaction, Community Functioning (see also the Appendix). Each topic, yielding a combination of questions from both the functional skills scale *and* the caregiver assistance scale, represented one important daily childhood activity. For example, the topic 'Mobility Outside' comprised questions of methods of outdoor locomotion, distance and speed of outdoor locomotion, outdoor surfaces, and car trans-

Table 1. Children included in the study.

JIA (N = 20)	Mono-articular (N= 2)	Oligo-articular (N= 10)	Poly-articular (N= 4)	Systemic articular (N= 4)
TI (N= 44)	A: Upper-extremity injury (N= 21)	B: Lower-extremity injury (N= 14)	Neurotrauma * (N= 9)	
NMD (N= 21)	Anterior Horn cell disease (N= 4)	Peripheral nerve disease (N= 4)	Muscular disease (N= 13)	

JIA = Juvenile Idiopathic Arthritis; TI = Traumatic Injury; NMD = Neuromuscular Disease; * Neurotrauma eventually in combination with A/B

fers. The clustering into topics enabled us to specify which changes in functional status occurred during the follow-up study.

Procedure

After being informed on matters all parents who participated in the study gave their consent. To improve validity, we administered the PEDI to the proximal caregiver. Interviews were administered at the onset of the disease in children with JIA and at consecutive follow-up visits at the hospital. The interviews followed after visiting the doctor and an assessment of a pediatric physiotherapist. All but two children with JIA were measured three times, two children were measured two times. Because symptoms may vary from day to day in children with JIA, we standardized the interviews by asking the parents to base their judgement on the past 14 days.
Parents of children with TI were interviewed within 14 days after the incident, and were asked to base their judgement on the functional status prior to the trauma as well as the 'actual' status. The children with TI were measured consecutively after three and seven months, so we collected data from four time periods.
All but three children with NMD, those with chronic conditions, were measured three times with an interval period of three and seven months respectively. The other three children with NMD were diagnosed with Guillain-Barré. The acute expression of this disease enabled us to collect data from the children before they became ill. We collected data from four time periods, as was done in children with TI.

Table 2. Age and interval periods in months.

	Age (mean) t1	**Mean time t1-t2**	**Mean time t2-t3**
TI (N = 44)	75.8 (SD = 44.5)	3.0 (SD = 0.4)	3.9 (SD = 0.6)
NMD (N = 21)	105 (SD = 35.3)	3.2 (SD = 0.4)	4.1 (SD = 0.6)
JIA (N = 20)	37.3 (SD = 22.3)	3.9 (SD = 1.8)	3.4 (SD = 1.2)

TI = Traumatic Injury; NMD = Neuromuscular Disease; JIA = Juvenile Idiopathic Arthritis

The mean age of the children in the three patient groups during the first interview (t_1) as well as the time-periods between the first and the second interview (t_2), and between the second and the third interview (t_3), are presented in Table 2.

Global rating scale

Before data collection, a global rating scale (GRS) was developed to predict changes in every child with respect to each of the nine clustered topics. The GRS served as an external standard in the responsiveness study. The baseline value in this GRS, always defined as 0, represented the PEDI outcome at first time point (t_1). Four senior clinicians rated the t_2, t_3, and when possible t_4, each of them in a relevant part of the clinical sample. The score modalities varied from value 0, -1, -2, to +1. Score 0 expressed no differences compared to the baseline situation. Score -1 represented a decrease in skills and/or increase in the amount of assistance for that particular topic compared to the baseline situation. Score -2 expressed a more severe functional limitation then score -1. In contrast, value +1 represented an increase in skills and/or decrease in amount of assistance for that topic compared to the baseline situation.
The GRS was fully completed at the same time of the first PEDI measurement in children with TI and NMD. Predictions made were based on the assumption that these children would follow a known course in functional status over time. The GRS in children with JIA was completed after three consultations by the pediatric physiotherapist, and predictions were made based on the results of each functional assessment.
The responsiveness of the PEDI topics are judged to be good when the sum scores on both the functional skills scale and caregiver assistance scale, with respect to the topic at hand, are in agreement with the global ratings.

Data Analysis

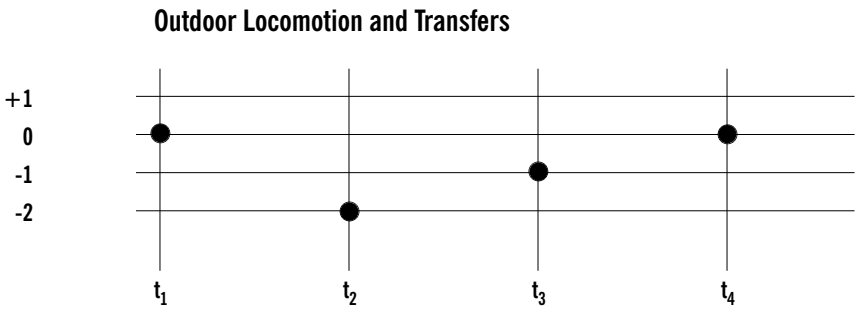
In this study, 'order restricted statistical inference'[25,26] was used to determine the responsiveness of the PEDI, for each combination of child and

topic. We want to explain 'order restricted statistical inference' as applied to our situation. Let X_1, X_2, X_3, and X_4 denote the scores of the PEDI topic at hand obtained at t_1, t_2, t_3, and t_4. The symbol M denotes the maximum score that can be obtained for that topic. At each time-point the score can be modeled using the binomial test model[27], i.e. X_t is a binomial random variable with success-probability P_t and M replications. Note that an implication of the binomial model is that P_t is the main determinant of X_t, i.e. the higher P_t the higher the score X_t.

Using this setup, the internal responsiveness[7] can be tested using the null hypothesis (H_0): $P_1=P_2=P_3=P_4$, i.e. the success-probabilities do not change across the time-points, and the alternative hypothesis (H_{1a}): P_1,P_2,P_3,P_4, i.e., the success-probabilities may differ between the time-points but the differences are not specified. In our study design we investigated the internal responsiveness[7] for each combination of child and topic in cases that no changes in outcome were expected over time. As a consequence, we would be satisfied if H_0 would be accepted (instead of rejected in favor of H_{1a}). Then the low variation of observed scores would be in accordance with the (constant) ratings on the GRS.

Furthermore, the external responsiveness[7] was investigated for each combination of child and topic in cases that changes in outcome were expected over time. A comparison would be made among the variation in the observed scores and the predictions obtained from the GRSs. To do that, H_{1a} was replaced by an alternative hypothesis that reflects the predictions.

An example, the GRS (e.g. topic outdoor locomotion and transfers) was filled in as follows:



The alternative hypothesis was then H_{1b}: $P_2 < P_3 < \{P_1, P_4\}$. This alternative hypothesis suggested the smallest success-probability at t_2, and the largest for t_1 and t_4. Note that various other alternative hypotheses (see Table 4) were derived from the GRSs for each combination of child and topic. In testing H_0 versus an order restricted H_1 hypothesis we would be satisfied if H_0 would be rejected.

A likelihood ratio test can be used to test H_0 versus an order restricted H_{1b}[25]. Since the exact distribution of this likelihood ratio test is unknown, a so-called 'plug-in' p-value will be computed[28]. To compute the likelihood ratio-test, estimates of the success-probabilities have to be obtained under H_0 (trivially easy, each probability is equal to $(X_1+X_2+X_3+X_4)/(4*M)$) and H_{1b}. The latter is done using the 'minimum violator algorithm, and simple extensions of this algorithm[25].

P-values were computed for each combination of child and topic. Values smaller than .2 indicate evidence in favor of the alternative hypothesis (either H1a or an order restricted alternative); values between .2 and .5 are indecisive; and, values larger than .5 indicate evidence in favor of the null-hypothesis. These thresholds are to some extent arbitrary. However, the main results were relatively independent of the actual threshold values (we also used threshold .10 and .40). Moreover, a better procedure to summarize these p-values is currently not available.

Results

Three hundred and ninety-six combinations of child and topic were rated in children with TI (44 children multiplied with nine topics). In 85.6% of the combinations, changes in functional status outcome over time were expected, i.e. H_1 hypothesis were used. Table 3 presents the percentages of the combinations in which changes were expected or not. As can be seen, most changes were expected to occur in children with TI. In contrast, the large number of H_0 hypothesis in the NMD group reflects the expectancy of a more stable outcome.

Table 4 presents the hypothesis, which was used in the study. Three kinds of H_0 hypothesis (stable outcome) and 16 kinds of order restricted H_1 hypothesis (changing outcome) were derived from the GRS scores. The order restricted H_1 hypothesis indicated improvement (hypothesis: 4, 5, 10, 11, 18), deterioration (hypothesis: 7, 8, 14, 16), or a combination of them. As one can notice, hypothesis 2, 3, 5, 8, 10, and 12 were used most frequently.

An overview of the p-values for each of three different groups of children is presented in Tables 5-a,b,c. Each table consists of two parts: for each topic a summary of the results obtained testing H_0 versus H_{1a}; and, a summary of the results obtained testing H_0 versus an order restricted alternative like H_{1b}. Considering topic 1 for the children with TI; no changes in functional status outcome were predicted in nine children (H_0: $t_1 = t_2 = t_3 = t_4$). The observed scores of eight children confirmed no evident changes, as their P-values were ≥ .5. Furthermore, significant changes were predicted in 35 children (e.g., H_1: $t_2 < t_1,t_3,t_4$). Seventeen children of them showed changes

Table 3. Percentages of hypothesis.

	H_0 hypothesis	H_1 hypothesis
TI (N=44)	14.4%	85.6%
NMD (N=21)	52.4%	47.6%
JIA (N=20)*	19.3%	80.7%

* one missing value topic 7-9; TI = Traumatic Injury; NMD = Neuromuscular Disease; JIA = Juvenile Idiopathic Arthritis

Table 4. Percentages of hypothesis.

Hypothesis		TI	NMD	JIA
H_0: $P_1=P_2$	1	-	-	4.0%
H_0: $P_1=P_2=P_3$	2	-	52.4%	15.3%
H_0: $P_1=P_2=P_3=P_4$	3	14.4%	-	-
H_1: $P_1<P_2$	4	-	-	6.2%
H_1: $\{P_1,P_2\}<P_3$	5	-	5.3%	30.5%
H_1: $P_2<\{P_1,P_3\}$	6	-	-	3.4%
H_1: $\{P_2,P_3\}< P_1$	7	-	9.5%	.6%
H_1: $P_3<\{P_1,P_2\}$	8	-	18.5%	3.4%
H_1: $\{P_1,P_3\}<P_2$	9	-	-	5.1%
H_1: $P_1<\{P_2,P_3\}$	10	-	-	31.6%
H_1: $\{P_1,P_2,P_3\}< P_4$	11	5.1%	-	-
H_1: $P_2<\{P_1,P_3,P_4\}$	12	63.6%	9.5%	-
H_1: $\{P_2,P_3\}<\{P_1,P_4\}$	13	7.1%	1.1%	-
H_1: $\{P_2,P_3,P_4\}<P_1$	14	2.0%	1.1%	-
H_1: $\{P_2,P_3\}<P_4< P_1$	15	2.3%	1.1%	-
H_1: $P_2<\{P_3,P_4\}<P_1$	16	.5%	.5%	-
H_1: $P_2<\{P_1,P_3\}<P_4$	17	1.3%	-	-
H_1: $\{P_1,P_2\}<\{P_3,P_4\}$	18	.3%	-	-
H_1: $P_2<P_3<\{P_1,P_4\}$	19	3.5%	1.1%	-

TI = Traumatic Injury; NMD = Neuromuscular Disease; JIA = Juvenile Idiopathic Arthritis

in a way as predicted, since their P-values were lesser than .2. Note that from eight children the P-values were between .2 and .5, representing moderate changes or changes, which were not predicted precisely.

As can be seen in Table 5-a, 82.4% (47/57) of the H_0 hypothesis, and 62.2% (211/339) of the H_1 hypothesis in the group children with TI were in accordance with the observed scores.

For the group NMD (see Table 5-b), these values were 95.9% for the H_0 hypothesis, and 38.8% for the H_1 hypothesis, respectively. Finally, the values for the group JIA (see Table 5-c) were 55.8% for the H_0 hypothesis, and 58.0% for the H_1 hypothesis.

Table 5-a. Results per topic in children with TI.

	H_0 versus H_{1a}				H_0 versus H_{1b}				
Topic	N	P< .2	.2 ≤ P < .5	P≥ .5	Topic	N	P< .2	.2 ≤ P < .5	P≥ .5
1	9	-	1	8	1	35	17	8	10
2	-	-	-	-	2	44	28	9	7
3	1	1	-	-	3	43	38	2	3
4	5	1	-	4	4	39	28	3	8
5	4	1	1	2	5	40	36	3	1
6	5	-	1	4	6	39	31	3	5
7	25	2	1	22	7	19	11	2	6
8	3	-	-	3	8	41	10	8	23
9	5	-	1	4	9	39	12	8	19
Total	**57**			**47**	total	**339**	**211**		

TI: Traumatic Injury

Table 5-b. Results per topic in children with NMD.

H_0 versus H_{1a}					H_0 versus H_{1b}				
Topic	N	P< .2	.2 ≤ P < .5	P≥ .5	Topic	N	P< .2	.2 ≤ P < .5	P≥ .5
1	15	-	-	15	1	6	5	1	-
2	6	-	-	6	2	15	5	6	4
3	6	-	-	6	3	15	5	3	7
4	14	-	1	13	4	7	5	-	2
5	7	-	-	7	5	14	6	4	4
6	6	-	-	6	6	15	4	2	9
7	14	-	3	11	7	7	2	3	2
8	17	-	-	17	8	4	1	-	3
9	14	-	-	14	9	7	2	-	5
Total	**99**			**95**	total	**90**	**35**		

NMD: Neuromuscular Disease

Table 5-c. Results per topic in children with JIA.

	H_0 versus H_{1a}				H_0 versus H_{1b}				
Topic	N	P< .2	.2 ≤ P < .5	P≥ .5	Topic	N	P< .2	.2 ≤ P < .5	P≥ .5
1	6	-	-	6	1	14	6	3	5
2	2	1	-	1	2	18	6	6	6
3	2	1	1	-	3	18	12	2	4
4	6	2	-	4	4	14	10	2	2
5	2	1	-	1	5	18	15	3	-
6	4	2	1	1	6	16	13	-	3
7	4	-	1	3	7	15	7	6	2
8	3	-	1	2	8	16	8	4	4
9	5	1	3	1	9	14	6	4	4
Total	**34**			**19**	total	**143**	**83**		

JIA: Juvenile Idiopathic Arthritis

The level of agreement between predictions and observed scores at topic level are shown in Table 6. When considering topic 1, 56.8% of the predictions (i.e., both H_0 hypothesis and H_1 hypothesis) that were made in children with TI, were in accordance with the observed scores. The percentages for the NMD and JIA group were 95.2% and 60%, respectively. It is remarkable that the percentages of agreement between predicted and observed outcome differ among the patient groups and the topics. Most agreement was found in children with NMD and TI.

Table 6. Agreement between predicted and observed outcome.

Topic		TI	NMD	JIA
Eating/drinking	1	56.8%	95.2%	60.0%
Grooming/bathing	2	63.6%	52.3%	35.0%
Dressing	3	86.3%	52.3%	60.0%
Toileting	4	72.7%	85.7%	70.0%
Indoor locomotion	5	86.3%	61.9%	80.0%
Outdoor locomotion	6	79.5%	47.6%	70.0%
Communication	7	75.0%	61.9%	52.6%
Play/interaction	8	29.5%	85.7%	52.6%
Community functioning	9	36.3%	76.1%	36.8%

TI = Traumatic Injury; NMD = Neuromuscular Disease; JIA = Juvenile Idiopathic Arthritis

Discussion

The purpose of this study was to examine the responsiveness of the Dutch PEDI. In review articles[2,6,7] there is consensus regarding the designs of responsiveness studies, particularly the need of an external standard which can support changes (or stability) in health as detected by the instrument. However, it is suggested that the most appropriate responsiveness statistic remains a matter of debate. Traditional approaches of responsiveness studies focus on the outcome at group-level[7]. In these approaches, even in a randomized clinical trial (which requires a homogeneous sample of subjects), one expects a similar outcome for each person. Variation between individuals is not (or partially) taken into account.

At the moment, there is an increasing interest in patient-centered outcome measures[29-31]. Each patient is unique, has individual concerns, and treatment effects can vary within individuals, even when receiving a similar treatment. Our approach was to investigate the responsiveness of the PEDI at an individual level. Analyses were performed for each combination of one child and one main topic of the PEDI.

It must be noted that the clustering of concurrent PEDI subscales into nine main topics is not in agreement with the way the instrument normally will be used for outcome measurement. However, this approach enabled us to specify the changes within a specific domain. For example, a child with a fractured leg has limitations in the selfcare domain. The scale score of the selfcare domain is an overall score and will not reflect whether limitations in the subscales 'using utensils during a meal', 'dressing pants', or 'toileting tasks' contribute to decreased scores.

A 'golden standard', which supports true changes, is mostly not available for validity or outcome studies. A global rating scale (GRS) was used in our study as the external standard. Predictions were made about the functional status development. As this might differ from child to child it implied that the responsiveness of each PEDI topic had to be determined for each child and that traditional methods for the assessment of responsiveness[7] could not be used. The parents did not fill in the GRSs because they were the respondents of the PEDI questionnaire. The four senior clinicians who scored the GRSs were regarded to serve as independent raters, as well as expert opinion deliverers.

In our study, we recruited children from three different patient groups. We supposed that they would show different courses in functional status over time. The first group is one of children with traumatic injuries. Limitations in functional status could be expected because of bone fractures, immobilization of the joints, pain or anxiety to fulfil daily activities. It was expected that their functional status level suddenly decreased after the injury, followed by a (partial) recovery within a few months. The assumption of changing levels explains the large number of H_1 hypothesis in Table 3, and more specifically the order restricted hypothesis 12 in Table 4. The analyses at topic level enabled us to differentiate between children with a singular arm and leg injury, as well as children with severe neurotrauma.

The second group contains children with a neuromuscular disease. All but three children were expected to have a chronic condition, and a functional status level which was stable or in decline over time. This is reflected by the large number of H_0 hypothesis in Table 3, and more specifically the order restricted hypothesis 8 in Table 4. Three children with Guillain Barré, however, were expected to follow a course like children with TI. Predictions about the functional status development in children with TI and NMD could be made based on outcome studies[32-36], and clinical experience.

In contrast, the course of functional status in children with JIA was supposed to be more influenced by day to day fluctuations in disease severity, affected joints, or pain that may occur. Therefore, the GRS was filled in after three assessments by the pediatric physiotherapist. The ratings were based on the results of a concurrent functional status measure: the Childhood Health Assessment Questionnaire (CHAQ)[37], and the joint assessment, reflecting the expectation of the expert opinion towards the PEDI topics. The large number of the order-restricted hypothesis 5 and 10 in Table 4 reflected the positive expectations with respect to the PEDI outcome over time. A majority of the children with JIA improved indeed with respect to the CHAQ scores and joint function when compared to the base line situation.

In our approach, the internal responsiveness or the ability of a measure to 'change' over a prespecified time frame[7] was tested using the H_0 hypothesis versus the H_{1a} hypothesis. The results reflected the agreement among the outcome of the PEDI topics and the ratings from the GRSs for these children in which functional status would be stable. It was expected that the

internal responsiveness be tested mainly in children with NMD. The large number of H_0 hypothesis that were accepted (Table 5-b) confirmed the ability of the PEDI to measure stability in functional status.

External responsiveness[7], testing the H_0 hypothesis versus a specified (order restricted) H_1 hypothesis, may even be more important because it reflects the extent to which changes in a measure relate to corresponding changes in a reference measure of functional status. These results reflected the agreement among the outcome of the PEDI topics and the ratings from the GRSs for these children in which functional status was changed. Around 60% of the H_0 hypothesis in children with TI and JIA were rejected (read: the alternative H_1 was accepted) (Table 5-a,c). As 16 alternative hypotheses were used, and each combination of child and topic was tested, we considered these results a conformation of the ability of the PEDI to measure specified changes in functional status in children with TI and JIA.

The results are less good for the children with NMD who were expected to deteriorate with respect to functional status (Table 4, hypothesis 7-8). Many children turned out to be stable or even improved. This regarded in particular the severely disabled children in whom the social context already had adapted to the child's limitations, and special equipment had been arranged (e.g., driving an electric wheelchair outside the house). The latter is illustrated in Table 6: the agreement between the GRSs and the PEDI scores in children with NMD with respect to topic 6 (i.e. outdoor locomotion) was moderate.

The different agreements (GRSs versus PEDI scores) at topic level (Table 6) may reflect a different responsiveness for each topic within a particular patient group. In children with TI, the topics 'Interaction' (topic 8) and 'community functioning' (topic 9) were not generally affected, although a deterioration was mostly rated on the GRSs. 'Dressing' (topic 3), 'toileting' (topic 4), and 'indoor/outdoor locomotion' (topic 5,6) on the contrary changed in a way as predicted. These findings need being addressed in future studies.

The results also demonstrate differences in agreement among the patient groups. The topic 'interaction' in children with NMD, for example, shows a high agreement between GRSs and PEDI scores, in contrast to the children with TI and JIA. This kind of analyses reveals that the responsiveness of the PEDI depends on the subjects and the topics of the PEDI which were measured.

The GRS was used as the external standard to support changes (or stability) as measured with the PEDI. The validity would be stronger when the GRS in children with TI and NMD was rated simultaneously with the PEDI assessment, i.e. after three and seven months, and the expert opinion was based on a clinical assessment. However, in most of the children with TI the medical treatment stopped within this time frame, and subsequently these children did not attend the hospital anymore. Moreover, the children with NMD visited the out-patients' department less frequently. Because the children with TI and NMD were supposed to have a known clinical course over time, the GRSs were filled in at baseline.

The GRSs in children with JIA were filled in retrospectively. The ratings were based on 'objective' parameters from the CHAQ and the joint assessment, and not on expert impression only, as in TI and NMD. These parameters were collected at the same time of the PEDI measurements, and they are thought to reflect the disease activity in JIA as well as health related functional status. Surprisingly, the overall agreement between GRSs and PEDI outcome are slightly better for children with NMD (68%) and TI (65%) than for children with JIA (57%). Whether the CHAQ and the joint assessment reflects the same aspects of functional status as the PEDI does, remains a question.

The choice of an external standard for responsiveness studies has to be made carefully. An external standard has limitations when it is based on predicted outcome. However, we argued that this approach could be used in children with known clinical courses. We agree with Husted et al.[7] that some individual variation in the external standard must be present for a study of external responsiveness to be sensible.

In summary, the responsiveness of the PEDI was examined at individual level using three different patient groups. Both internal responsiveness and external responsiveness were tested using 'order restricted statistical inference'. A GRS was used as the external standard to support changes (or stability) in outcome. The internal responsiveness of the PEDI varies from moderate in children with JIA to excellent in children with TI and NMD. The external responsiveness varies from moderate in children with NMD, to good in children with TI and JIA. The agreement between GRS and PEDI differs for each topic and patient group. The overall agreement is more demonstrated in patient groups with a known clinical course, indicating a

useful concept in measuring responsiveness of new instruments. These study results support the author's opinion that the Dutch PEDI is able to measure changes in functional status over time. Finally, more research is needed to refine this approach in order to improve responsiveness studies, in patient-centered health outcome.

Appendix Chapter 6

Content of PEDI topics in the study:

1) **Eating/Drinking:**
   Types of food textures
   Use of utensils
   Use of drinking containers
   Amount of caregiver assistance

2) **Grooming/Bathing:**
   Tooth brushing
   Hair brushing
   Nose Care
   Hand washing
   Washing body and face
   Amount of caregiver assistance

3) **Dressing:**
   Pullover/front-opening garments
   Fasteners
   Pants
   Shoes/socks
   Amount of caregiver assistance

4) **Toileting:**
   Toileting tasks
   Management of bladder
   Management of bowel
   Amount of caregiver assistance

5) **Indoor Locomotion and Transfers:**
   Toilet transfers
   Chair/wheelchair transfers
   Bed transfers
   Tub transfers

Indoor locomotion-methods
Distance/speed indoors
Pulls/carriers objects
Upstairs
Downstairs
Amount of caregiver assistance

6) **Outdoor Locomotion and Transfers:**
Outdoor locomotion- methods
Distance/speed outdoors
Outdoor surfaces
Car transfers
Amount of caregiver assistance

7) **Communication:**
Comprehension of word meanings
Comprehension of sentence complexity
Functional use of communication
Complexity of expressive communication
Amount of caregiver assistance

8) **Play and Interaction:**
Problem-resolution
Social interactive play
Peer interactions
Play with objects
Amount of caregiver assistance

9) **Community Functioning:**
Self-information
Time orientation
Household chores
Self-protection
Community function
Amount of caregiver assistance

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Summary
General Discussion
Conclusions

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Chapter 7

Summary

In Chapter 1 the theoretical concept of childhood disablement is explained. In addition, a brief introduction is presented, regarding pediatric functional status measurement, and more specifically the subject of this thesis: the Pediatric Evaluation of Disability Inventory. The aims and outline of the thesis are formulated.

In Chapter 2 the applicability of the PEDI in the Netherlands is studied involving a sample of 20 Dutch children without disabilities. Their functional status outcome is compared to American peers; the Rasch statistical model was used to analyze the Dutch data. Score profiles were found not to be compatible with the score profiles of American children. More specifically, the data suggested inter-cultural differences. Based on this study, it is argued that cross-cultural validation of the PEDI was necessary before using it in the Netherlands.

The translation and adaptation procedure in developing a Dutch version of the PEDI was based on current scientific guidelines in cross-cultural research as outlined in Chapter 3. A content validity study of the Dutch PEDI was subsequently performed and is also presented in this chapter. In this study, 31 allied health professionals completed a validity questionnaire. They confirmed the functional content and feasibility of the Dutch PEDI for pediatric functional status measurement. Both the adaptation process and content validity study generated the addition of eight new questions into the Dutch PEDI. Four of them are linked to an essential daily childhood activity in the Netherlands: 'bicycling'.

In Chapter 4 the reliability of the Dutch PEDI is examined. Therefore, parents of 64 children without disabilities and parents of 31 children with disabilities were interviewed. Inter-interviewer reliability was studied after scoring audio-taped interviews by a second researcher. For intra-respondent reliability the same parent was interviewed twice within two weeks; for inter-respondent reliability both parents of a child were interviewed independently within a few days. The proportion of matching scores varied from

good to excellent in inter-interviewer reliability and inter-respondent reliability, and from moderate to excellent in intra-respondent reliability. Intraclass correlation coefficients were all excellent. Cronbach's alpha (internal consistency) was calculated on a sample of 63 healthy children aged two years, and varied from excellent in the self-care and social function domain, to moderate in the mobility domain. Although some adaptations have to be made, good psychometric properties of the Dutch PEDI have been established.

In Chapter 5 the discriminative validity of the Dutch PEDI is examined in 62 children without disabilities and in 197 children with disabilities (Infantile Encephalopathy, N= 40; Juvenile Idiopathic Arthritis, N= 20; Neurometabolic conditions, N=36; Neuromuscular Disorders, N= 9; Skeletal Disorders, N= 28; Spina Bifida, N= 41; Traumatic Injury, N= 23). Discriminant analysis revealed an excellent degree of sensitivity and specificity of the Dutch PEDI in identifying childhood disablement (non-disabled vs. disabled). The results also suggest the identification of subsets within the clinical samples: functional status linked to CNS involvement and musculo-skeletal involvement indicating potentials of the Dutch PEDI to discriminate within specific clinical groups.

In Chapter 6 the responsiveness of the Dutch PEDI was examined. In contrast to traditional approaches, the responsiveness was measured at individual level. Children with Traumatic Injury (TI), Neuromuscular Diseases (NMD), and Juvenile Idiopathic Arthritis (JIA) (N=85) were measured three times. A Global Rating Scale (GRS) was used as an external standard of change; analyses were done at an individual level. Internal and external responsiveness were tested using 'order restricted statistical inference'. The internal responsiveness of the PEDI varied from moderate to excellent. The external responsiveness varied from moderate to good. The study results support the author's opinion that the Dutch PEDI is able to measure meaningful individual changes in functional status over time.

General discussion

Whether a child has functional deficits in daily living activities should be of interest to all health professionals in pediatric (re)habilitation and pediatrics. 'An advantage of tracking functional status is that decline in performance warrants comprehensive evaluations for biomedical conditions that interfere with developmental competencies'[1]. The (Dutch) PEDI can thereby serve as a diagnostic tool and can be used for program evaluation in pediatric (re)habilitation medicine. Moreover, it can serve as an outcome instrument for medical/surgical treatment[2-5]. 'By describing the complexity of genetic mechanisms on developmental and functional status, medical professionals can optimize quality of life, provide family support, and critically contribute knowledge to preventive efforts'[1].

Two aspects of childhood disability became increasingly important throughout the last decade:

1. The focus on the child's physical limitations has been changed into a broader assessment of family dynamics, needs, resources, and coping-style mechanisms[6,7], and
2. The environmental demands and factors[8].

The role of the parents in childhood disability and pediatric (re)habilitation programs is reviewed by Ketelaar et al.[9]. They recommended an active role of parents in all phases of a therapeutic program, such as goal setting, and they underlined the task of the therapist to 'support the naturally occurring opportunities that exist at home to learn and practice daily skills'. These skills should be linked to the child's contextual demands and supports in his/her relevant environment. Therefore, context-specific functional testing of children need to be incorporated into physical therapy practice[8]. Functional assessment using the (Dutch) PEDI enables the researcher to describe the child's strengths and challenges in the context of essential activities within the child's everyday environment.
Recently, the PEDI proved to be an accurate evaluative outcome measure for function focussed physical therapy in children with Cerebral Palsy[10]. Main features of this new approach in pediatric physical therapy are the

establishment of concrete functional goals, treatment of functional skills in a relevant environment, an active role of the child (problem solving), and an active parental involvement (goal setting, implementation in daily life)[9]. The PEDI's concept corresponds to current developments in pediatric physical therapy and (re)habilitation. The main goal of treatment in these settings regarding children with developmental deficits is to optimize childhood daily functioning and minimize caregiver assistance.

This thesis focuses on the applicability and psychometric properties of the (Dutch) PEDI.
Although in Chapter 2 the study sample was small, the study findings demonstrated outcome differences between the American normative sample and the Dutch study sample indicating cultural influences. Because the particular items (which caused deviant fit-scores) are relevant for both cultures, and the conceptual equivalence is confirmed (Chapter 3), the study results of Chapter 2 require a new calibration of the PEDI for the use in the Netherlands. This calibration is planned using large samples of children without disabilities in the age range of 6 months to 7.5 years. The PEDI outcome from a Dutch normative sample enables the researcher to compare item-difficulty cross-culturally according to the standard approach for cross-cultural research[11,12].

Adaptations were made to improve the validity and applicability of the PEDI for use in the Netherlands. Until now, little research has been done to delineate what is essential from what is supplementary in the process of cross-cultural adaptation[13]. The adaptation procedure followed in this thesis was based on scientific guidelines[13-15]. Recently, Perneger[16] has addressed the current lack of empirical evidence about the effectiveness of various procedures used for translating psychometric instruments. More research is needed to determine accurate translation and adaptation methods in realizing outcome measures for cross-cultural usage.
The advantage of the procedure followed (Chapter 3) is the participation of a multi-disciplinary team which secures a) the broad aspects of childhood disability in the Netherlands, b) the accuracy of the translation, and c) methodological improvements in the development of the Dutch PEDI.

Reliability of the Dutch PEDI is confirmed in this thesis (Chapter 4). With regard to the internal consistency it has yet to be examined for other age groups as well. With regard to the other aspects of reliability, test-time might be a factor that can partially explain the disagreement found in inter-interviewer, inter-respondent, and intra-respondent reliability. The disadvantage of the test-time is also mentioned by the respondents in the content validity study (Chapter 3). The administration of the complete PEDI takes 45-60 minutes and it seems hard to be fully concentrated all that time. For clinical use it is not always indicated to complete the whole interview. Test-time can be reduced by selecting a target scale, e.g. the functional skills scale and caregiver assistance scale of the mobility domain; each PEDI scale is self-contained and can be used separately. The development of short forms of the PEDI for each age class will also contribute to shorten the test-length in future.
The parent's agreement of their child's functional status was not studied before. The reliability study in this thesis reveals parental differences. More specifically, mothers judged their children more capable and less dependent on their assistance than fathers did, especially when the child is disabled. Because the PEDI is a judgment-based outcome instrument it is strongly recommended to interview the same parent when the PEDI is used for evaluative purposes.

An important challenge in pediatric (re)habilitation research is the use of the Rasch rating scale methodology. This statistical approach, based on the Item Response Theory, enables researchers to develop disease related functional status profiles in the future. It is conceivable that the item-difficulty for skills regarding bladder management will be different in children with Spina Bifida compared to children with Juvenile Idiopathic Arthritis. An appropriate disease-related 'standard' in functional status assessment is preferable. 'Treatment strategies should focus primarily at improving functional ability respecting the typical characteristics of the disease and the disease related natural course'[17]. It facilitates investigating the individual needs of a child compared to his/her peers with the same disease.
At the moment, the Dutch PEDI is able to discriminate between children with and without disabilities, and the study findings in Chapter 5 support the potentials of the Dutch PEDI to discriminate between children with Central

Nervous System (CNS) involvement and children with musculo-skeletal involvement. Recently, these study findings were supported by Haley et al.[18].

The most appropriate statistical and methodological approach in measuring responsiveness of health-related outcome instruments has up to now been a matter of debate[19]. Our approach to examine the responsiveness of the Dutch PEDI at an individual level is in line with current developments, in which it is increasingly important to aim at patient-centered health services[20] and to take a patient-centered approach to outcome measurement[21,22]. The responsiveness of the Dutch PEDI is confirmed in this thesis. However, future research is needed to refine and establish this kind of responsiveness in outcome measurement studies.

Conclusions

The following conclusions can be drawn from this thesis:

1. The original PEDI, both the content and the reference values, cannot be simply applied for functional status assessment in the Netherlands.

2. The adaptation process and the results from the content validity study resulted in the addition of eight new items; four of them are linked to a typical Dutch essential daily activity for the PEDI: 'bicycling'.

3. A content validity study confirmed the functional content and feasibility of the Dutch adapted PEDI for pediatric functional status assessment in the Netherlands.

4. The reliability of the Dutch PEDI is confirmed in this thesis.

5. The Dutch PEDI discriminates excellently between children with and without functional deficits regarding childhood daily activities.

6. The Dutch PEDI is able to measure individual changes in functional status over time.

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Nederlandse samenvatting

In Hoofdstuk 1 wordt een inleiding gegeven over het 'disablement process'. De gevolgen van aangeboren aandoeningen en ziekten voor het (bewegend) functioneren worden in een model weergegeven waarbij onderscheid wordt gemaakt in stoornissen, beperkingen van activiteiten uit het dagelijkse leven en beperkingen van functioneren in de samenleving. Bij kinderen is dit model meer gecompliceerd omdat groei en ontwikkeling, alsmede de rol van ouders van invloed zijn op het dagelijks functioneren.
Adequate meetinstrumenten zijn nodig om op elk nivo van dit model de gevolgen van aangeboren aandoeningen en ziekten op het (bewegend) functioneren te beschrijven. In dit Hoofdstuk wordt een inleiding gegeven over meetinstrumenten en de eisen die gesteld worden aan een meetinstrument voor kinderen. Er wordt specifiek ingegaan op het hoofdthema van dit proefschrift: de 'Pediatric Evaluation of Disability Inventory' (PEDI).
De PEDI is een gestructureerde vragenlijst waarmee beperkingen kunnen worden gemeten van activiteiten uit het dagelijkse leven van kinderen in de leeftijd van 6 maanden tot 7.5 jaar. De vragenlijst is gericht op drie domeinen: zelfverzorging, ambulantie en sociaal functioneren.
De invloed van groei en ontwikkeling op het dagelijks functioneren van een kind is een belangrijke pijler in het theoretisch construct van de PEDI. Er zijn Amerikaanse normwaarden voor 14 leeftijdsklassen (van elk 6 maanden). Voorbereidingen zijn gestart voor een normeringsstudie in Nederland.
Een andere belangrijke pijler in het theoretisch construct van de PEDI is dat zowel vaardigheden van het kind kunnen worden gemeten alsook de context waarin het kind functioneert. Omgevingsfactoren zoals de rol van ouders of de inrichting van de leefomgeving zijn van invloed op het dagelijks functioneren. Dit proefschrift beschrijft de ontwikkeling van de Nederlandse versie van de PEDI. Het doel, de vraagstellingen en de inhoud van het proefschrift worden tenslotte in dit Hoofdstuk uiteengezet.

In Hoofdstuk 2 van dit proefschrift worden de resultaten beschreven van de bruikbaarheid van de Amerikaanse versie van de PEDI in Nederland. In deze studie werd de PEDI afgenomen bij de ouders van 20 gezonde kinderen. De resultaten werden vergeleken met de normwaarden van

Amerikaanse kinderen. Fit-scores blijken bij meer dan 75% van de kinderen afwijkend in het domein sociaal functioneren. Met behulp van 'person fit' analyses (volgens het Rasch model) werd van elk item uit de vragenlijst berekend hoe groot de kans zou zijn dat een kind een item positief scoort op basis van de totale score van dat kind. De voorspelde kans werd vervolgens vergeleken met de geobserveerde waarde. Op deze manier konden een aantal items worden getraceerd die anders gescoord werden door de Nederlandse kinderen in vergelijking tot de Amerikaanse normgroep.
Op basis van deze onderzoeksgegevens wordt geconcludeerd dat er sterke aanwijzingen zijn voor culturele verschillen in het dagelijks functioneren van kinderen in Nederland en Noord-Amerika en dat de PEDI derhalve aangepast en opnieuw gecalibreerd dient te worden.

In Hoofdstuk 3 is de Nederlandse bewerking van de PEDI beschreven. Dit proces was gebaseerd op wetenschappelijke richtlijnen afkomstig uit 'cross-cultural research'. Allereerst werd de PEDI in het Nederlands vertaald door een gecertificeerd vertaler. Vervolgens werden inhoudelijke en tekstuele aanpassingen gemaakt, waarna de Nederlandse vragenlijst werd 'terugvertaald' in het Amerikaans door een vertaler van Noord-Amerikaanse afkomst. De aanpassingen werden verricht door een multi-disciplinair team met werkervaring in de begeleiding van kinderen met ontwikkelingsstoornissen. De volgende twee criteria werden aangehouden tijdens het adaptatieproces: 1) elk item werd onderzocht op relevantie en 2) elk item werd na de vertaling gecheckt of de essentie van de vraag bewaard was gebleven. Uiteindelijk werden in de vaardighedenschaal 4 items toegevoegd aan het geheel van 197 items.
Een onderzoek naar inhoudsvaliditeit werd gestart na de bewerkingsfase. Inhoudsvaliditeit kan beschouwd worden als een index waarmee aangetoond kan worden dat de items uit de PEDI en het instrument in zijn geheel het construct representeren, dat het beoogt te willen meten. Een panel bestaande uit 32 professionals werkzaam in de jeugdgezondheidszorg werd benaderd voor het invullen van een enquete over de PEDI. Eenendertig respondenten retourneerden een ingevulde enquete. Samengevat bevestigen zij dat de inhoud van de PEDI representatief en bruikbaar is voor het meten van 'disability' bij kinderen in de genoemde leeftijdscategorie. Desondanks werden, naar aanleiding van de reakties van de respondenten,

4 extra vragen ingepast in de uiteindelijke Nederlandse PEDI. Deze vragen gaan over de vaardigheid fietsen.

In Hoofdstuk 4 werd de betrouwbaarheid van de Nederlandse PEDI onderzocht. Betrouwbaarheid van een meting heeft te maken met de reproduceerbaarheid van een meting. Een meting wordt onbetrouwbaar genoemd wanneer 1) steeds andere waarden worden gevonden bij herhaalde metingen of 2) verschillende waarden worden gevonden tijdens afname door verschillende personen. Bij het onderzoek naar de overeenkomst tussen de scores bij herhaalde metingen (test-hertest betrouwbaarheid) werd dezelfde ouder geïnterviewd na 14 dagen. Bij het onderzoek naar de overeenkomst tussen de scores van verschillende interviewers (inter-interviewer betrouwbaarheid) werd gebruik gemaakt van een audio-tape. Bij het onderzoek naar de overeenkomst tussen de scores van beide ouders (inter-respondent betrouwbaarheid) werden beide ouders geïnterviewd binnen enkele dagen. Zij werden gevraagd niet tussentijds over het interview te praten. In deze studie namen 63 gezonde 2-jarige kinderen en ouders deel en 31 kinderen en ouders die bekend zijn in het Universitair Medisch Centrum/WKZ te Utrecht met beperkingen in het dagelijks functioneren.
De proportie overeenkomstige scores varieert van .43 tot 1.0 voor de testhertest betrouwbaarheid, .81 tot 1.0 voor de inter-interviewer betrouwbaarheid en van .65 tot 1.0 voor de inter-respondent betrouwbaarheid. Een andere maat voor de betrouwbaarheid, de Intra-Class Correlatie Coëfficient is op één na boven de .90. Tenslotte werd nagegaan hoe de items uit de PEDI onderling samenhangen (Interne consistentie). De maat daarvoor, Cronbach's alfa is .89 voor het domein zelfverzorging, .48 voor het domein ambulantie en .87 voor het domein sociaal functioneren. Toekomstig onderzoek is nodig om de interne consistentie te meten bij andere leeftijdsgroepen.Op basis van dit onderzoek werden 'slecht scorende' items geëvalueerd en zonodig aangepast en wordt geadviseerd om follow-up metingen te verrichten bij dezelfde ouder.

In Hoofdstuk 5 is het discriminatieve vermogen van de NL PEDI onderzocht. In deze studie wordt onder discriminatief vermogen verstaan 'het vermogen om onderscheid te maken tussen kinderen met en zonder beperkingen in dagelijks functioneren'. Hiertoe werden 62 gezonde kinderen ver-

geleken met 197 kinderen die bekend zijn in het Universitair Medisch Centrum/WKZ te Utrecht. De laatste groep bestond uit 40 kinderen met een Infantiele Encefalopathie, 20 kinderen met een Juveniele Idiopathische Artritis, 36 kinderen met een psychomotore achterstand die voor diagnostiek werden opgenomen op de afdeling neuro-metabole ziekten, 9 kinderen met een neuromusculaire aandoening, 28 kinderen met Osteogenesis Imperfecta, 41 kinderen met Spina Bifida en 23 kinderen met traumatisch letsel. Door middel van discriminant analyse werd aan de hand van het scorepatroon van ieder kind een voorspelling gedaan van welke groep het kind afkomstig is. Van de 62 kinderen zonder beperkingen werd 94% goed voorspeld. Van de 197 kinderen met beperkingen in dagelijks functioneren werd 92% voorspeld dat ze afkomstig zijn uit een van de klinische groepen. Binnen de klinische groepen variëerden de percentages van 21% tot 75% afhankelijk van de diagnose. De resultaten geven een indicatie dat de Nederlandse PEDI onderscheid kan maken tussen kinderen met centraal neurologische aandoeningen en kinderen met primair aandoeningen van het bewegingsapparaat.

In Hoofdstuk 6 is het vermogen van de Nederlandse PEDI onderzocht om veranderingen in functioneren te kunnen meten. Hiertoe werden 85 kinderen op 3 tijdstippen gemeten. Er waren 20 kinderen met Juveniele Idiopathische Artritis, 44 kinderen met traumatisch letsel en 21 kinderen met een neuromusculaire aandoening. Deze patientengroepen werden geselecteerd omdat verwacht werd dat zij met betrekking tot het dagelijks functioneren elk op een andere wijze zouden veranderen. Op een schaal van -2 tot +1 werd door een onafhankelijk deskundige gescoord óf en op welke wijze een verandering te verwachten viel op een van de onderdelen van de Nederlandse PEDI. Deze voorspelling werd vergeleken met de geobserveerde scores van de Nederlandse PEDI.
De analyse werd gedaan met behulp van ‘order restricted statistical inference’. Deze techniek is niet gebruikelijk in ‘responsiveness’ studies. Traditionele studies vergelijken de uitkomstmaat van groepen patiënten. In onze studie werden analyses gedaan waarbij van elk kind werd nagegaan of de verwachte verandering bij een kind overeenkomt met de PEDI score. De meeste veranderingen in functioneren werden verwacht bij kinderen met JIA en TI. De mate van overeenkomst tussen voorspelde en geobserveerde

waarden variëerde van matig tot uitstekend en bleek afhankelijk van patientengroep en PEDI topic. Toekomstig onderzoek zal deze manier van responsiviteitsstudies verder moeten verfijnen.

Het concept van de Nederlandse PEDI past in de visie om breder te kijken dan alleen naar de beperkingen van het kind. Omgevingsfactoren zoals de rol van ouders, het dynamisch proces binnen het gezin, de eisen die gesteld worden van buitenaf en de inrichting van de omgeving zijn minstens zo belangrijk om over het dagelijks functioneren van een kind tot een goed oordeel te komen.
Voor alle hulpverleners in de kinderfysiotherapie, de kinderrevalidatie en de kindergeneeskunde is de vraag relevant of het kind beperkingen heeft in dagelijks functioneren. De Nederlandse PEDI is een meetinstrument wat daarvoor gebruikt kan worden.

Op basis van de onderzoeksgegevens in dit proefschrift kunnen de volgende conclusies worden getrokken:

1. De inhoud en de normwaarden van de Amerikaanse PEDI kunnen niet zonder meer worden toegepast/gebruikt voor onderzoek naar dagelijkse vaardigheden bij kinderen in Nederland.
2. Het adaptatieproces resulteerde in het toevoegen van 4 nieuwe items. Naar aanleiding van de onderzoeksresultaten naar de 'inhouds validiteit' werden nog eens 4 items toegevoegd. Deze items gaan over een voor Nederlandse begrippen essentiele dagelijkse vaardigheid: fietsen.
3. De onderzoeksresultaten van de studie naar de 'inhouds validiteit' bevestigen de representativiteit en bruikbaarheid van de Nederlandse PEDI voor onderzoek naar 'disability' bij kinderen.
4. De meeste vormen van betrouwbaarheid van de Nederlandse PEDI zijn onderzocht en bleken te variëren van matig tot uitstekend.
5. De Nederlandse PEDI discrimineert uitstekend tussen kinderen met en zonder beperkingen in dagelijks functioneren.
6. De Nederlandse PEDI is in staat individuele veranderingen in de tijd te meten met betrekking tot het nivo van dagelijkse vaardigheden bij kinderen.

Appendix: Score-formulier research-versie PEDI

Informatie over het kind:

Naam: _____
Geslacht: M ❒ V ❒ Ethnische afkomst: _____

Leeftijd	dag	maand	jaar
interviewdatum:	_____	_____	_____
geboortedatum:	_____	_____	_____
leeftijd:	_____	_____	_____

Onderzoeksgroep: _____
Patiëntnummer: _____
Diagnose: _____

Huidige situatie van het kind:

❒ woont thuis
❒ opname in zorginstelling: _____
❒ anders (noteer): _____
School of andere faciliteit: _____

Informatie over de respondent:

Naam: _____
Geslacht: M ❒ V ❒
Relatie tot het kind: _____
Beroep: _____
Opleiding: _____
Verzorging: ❒ <25% ❒ <50% ❒ ≥50% ❒ >75%

Informatie over de interviewer:

Naam: _____
Relatie tot onderzoek: _____

Informatie over het onderzoek:

Afname: ❒ thuis ❒ WKZ ❒ anders _____
Methode: ❒ interview ❒ interview per telefoon ❒ schriftelijk
❒ Pretest ❒ inter-respondent betrouwbaarheid ❒ verzorger-verzorger
❒ verzorger-hulpverlener
❒ inter-tester betrouwbaarheid
❒ intra-tester betrouwbaarheid
❒ interne consistentie

❒ Discriminant validiteit ❒ Responsiveness-to-change
❒ Construct validiteit ❒ Normeringsonderzoek

Deel 1: Vaardighedenschaal
Zelfverzorging

A. Soorten Voedselstructuren	*0*	*1*	*E. Haarkammen*	*0*	*1*
1. Eet gepureerd voedsel			21. Houdt hoofd in positie		
2. Eet geprakt voedsel			22. Brengt borstel of kam naar haar		
3. Eet gesneden voedsel			23. Borstelt of kamt haar		
4. Eet alle soorten voedselstructuren			24. Maakt scheiding en ontklit		
***B.** Gebruik van bestek*	*0*	*1*	***F.** Verzorging neus*	*0*	*1*
5. Eet met vingers			25. Staat afvegen van de neus toe		
6. Schept met lepel en brengt naar mond			26. Poogt neus te snuiten		
7. Gebruikt lepel goed			27. Veegt neus af op verzoek		
8. Gebruikt vork goed			28. Veegt neus af op eigen initiatief		
9. Gebruikt mes om te smeren, snijdt zacht voedsel			Snuit en veegt de neus af op eigen initiatief		
10. Gebruikt bestek samen			***G.** Handenwassen*	*0*	*1*
***C.** Gebruik van drinkgerei*	*0*	*1*	30. Steekt handen uit om te wassen		
11. Houdt flesje of tuitbeker vast			31. Wrijft handen tijdens wassen		
12. Brengt beker naar de mond, beker kiept			32. Hanteert waterkraan, pakt zeep		
13. Tilt beker op, met 2 handen			33. Wast handen grondig		
14. Tilt beker op, met 1 hand			34. Droogt handen grondig		
15. Schenkt drinken uit pak of kan			***H.** Lichaam en gezicht wassen*	*0*	*1*
***D.** Tandenpoetsen*	*0*	*1*	35. Poogt delen lichaam te wassen		
16. Houdt mond open bij tandenpoetsen			36. Wast lichaam grondig; gezicht niet		
17. Houdt zelf tandenborstel vast			37. Pakt zeep (en gebruikt washand, indien van toepassing)		
18. Poetst tanden; niet grondig			38. Droogt lichaam grondig		
19. Poetst tanden; grondig			39. Wast en droogt gezicht grondig		
20. Brengt tandpasta op borstel					

I. Kleding bovenlichaam	*0*	*1*
40. Helpt, zoals steekt arm door mouw		
41. Trekt t-shirt, jurk of trui uit (zonder sluitingen)		
42. Trekt t-shirt, jurk of trui aan (zonder sluitingen)		
43. Trekt voor-sluitende kleding aan en uit (zonder sluitingen)		
44. Trekt voor-sluitende kleding aan en uit inclusief sluitingen		
J. Sluitingen	*0*	*1*
45. Helpt met sluitingen		
46. Ritst open en dicht		
47. Drukknoopjes open en dicht		
48. Knopen open en dicht		
49. Ritst open en dicht; deelt en haakt rits vast		
K. Broeken	*0*	*1*
50. Helpt, zoals duwt benen door broeks-pijp		
51. Trekt broek uit, elastische taille		
52. Trekt broek aan, elastische taille		
53. Trekt broek uit, inclusief losmaken sluitingen		
54. Tekt broek aan, inclusief vastmaken sluitingen		
L. Schoenen/sokken	*0*	*1*
55. Trekt sokken en schoenen uit; veters los		
56. Trekt schoenen aan; veters los		
57. Trekt sokken aan		
58.trekt schoenen aan; juiste voet		
59. Strikt veters		

M. Toiletvaardigheden	*0*	*1*
60. Helpt met aan en uitkleden		
61. Poogt af te vegen na toilet		
62. Hanteert toiletbril, toiletpapier en spoelt toilet door		
63. Kan aan en uitkleden tijdens toiletgang		
64. Veegt zelf goed af (ontlasting)		
N. Blaasfunctiecontrole	*0*	*1*
65. Geeft natte luier aan		
66. Meldt af en toe wanneer het moet plassen (overdag)		
67. Meldt consequent tijdig wanneer het moet plassen (overdag)		
68. Onderneemt zelf actie om naar toilet te gaan (overdag)		
69. Blijft consequent droog (dag en nacht)		
O. Darmfunctiecontrole	*0*	*1*
70. Meldt wanneer het verschoond moet worden (ontlasting)		
71. Meldt af en toe wanneer het moet poepen (overdag)		
72. Meldt consequent tijdig wanneer het moet poepen (overdag)		
73. Maakt onderscheid tussen aandrang plassen en poepen		
74. Gaat zelf naar toilet om te poepen, geen "ongelukjes" meer		
Somscore Zelfverzorgingsdomein		
Alle items ingevuld?		
Opmerkingen		

Deel 1: Vaardighedenschaal
Verplaatsen

A. Toilettransfers	*0*	*1*	18. Komt tot zitten op rand van bed; gaat liggen vanuit zithouding		
1. Zit gesteund door voorziening of verzorger			19. Gaat in en uit eigen bed		
2. Zit ongesteund op kinderpotje of toilet			20. Gaatin en uit eigen bed, armsteun niet noodzakelijk		
3. Gaat op en af laag toilet of kinderpotje			***E. Badtransfer***	*0*	*1*
4. Gaat op en af toilet voor volwassene			21. Zit gesteund door voorziening of verzorger		
5. Gaat op en af toilet, armsteun niet noodzakelijk			22. Zit ongesteund en verplaatst in bad/douche		
B. Stoel/ Rolstoeltransfers	*0*	*1*	23. Klimt of schuift op en af rand van bad/douche		
6. Zit gesteund door voorziening of verzorger			24. Gaat zitten en staat op in bad/douche		
7. Zit ongesteund op stoel of bank			25. Stapt/maakt transfer in en uit bad/douche		
8. Gaat op en af laag stoeltje of meubel			***F. Wijze verplaatsen binnenshuis***	*0*	*1*
9. Gaat in en uit stoel voor volwassene of eigen rolstoel			26. Rolt, schuift, kruipt over vloer		
10. Gaat in en uit stoel, armsteun niet noodzakelijk			27. Loopt, steun noodzakelijk (meubel, muur, verzorger, hulpmiddel)		
C. Autotransfers	*0*	*1*	28. Loopt zonder steun		
11. Verplaatst in auto; schuift in en gaat in en uit autostoeltje			***G. Afstand en snelheid binnenshuis***	*0*	*1*
12. Gaat in en uit auto met weinig hulp of aanwijzingen			29. Verplaatst binnen 1 vertrek; moeizaam (valt, langzaam voor leeftijd)		
13. Gaat in en uit auto zonder hulp of aanwijzingen			30. Verplaatst binnen 1 vertrek; zonder moeite		
14. Hanteert veiligheidsgordel of stoelfixatie			31. Verplaatst tussen vertrekken; moeizaam (valt, langzaam voor leeftijd)		
15. Gaat in en uit auto en opent en sluit portier			32. Verplaatst tussen vertrekken; zonder moeite		
D. Bedtransfer	*0*	*1*	33. Verplaatst 15 meter; opent en sluit binnen en buitendeuren		
16. Verandert van houding in bed of ledikant					
17. Komt tot zitten in bed of ledikant					

H. Voorwerpen duwen/dragen	*0*	*1*	*L. Fietsen*	*0*	*1*
34. Verandert doelbewust van plaats			52. Fietst op (aangepaste) driewieler		
35. Verplaatst voorwerpen over vloer			53. Fietst op tweewieler met zijwieltjes (10 m)		
36. Draagt kleine voorwerpen; 1 hand			54. Fietst op tweewieler met hulp (50 m)		
37. Draagt grote voorwerpen; 2 handen			55. Fietst op tweewieler zonder hulp (100 m)		
38. Draagt breekbare/te morsen voorwerpen					
I. Wijze verplaatsen buitenshuis	*0*	*1*	*M. Trap op gaan*	*0*	*1*
39. Loopt, steun noodzakelijk (voorwerpen, verzorger, hulpmiddel)			56. Schuift of kruipt gedeelte van trap op (1 - 11 treden)		
40. Loopt zonder steun			57. Schuif of kruipt gehele trap op (12 – 15 treden)		
41. Loopt en draagt voorwerp			58. Loopt gedeelte van trap op		
			59. Loopt gehele trap op; met moeite (langzaam voor leeftijd)		
			60. Loopt gehele trap op; zonder moeite		
J. Afstand en snelheid buitenshuis	*0*	*1*	*N. Trap af gaan*	*0*	*1*
42. Verplaatst 3 – 15 meter			61. Schuift of kruipt gedeelte van trap af (1 – 11 treden)		
43. Verplaatst 15 – 30 meter			62. Schuift of kruipt gehele trap af (12 –15 treden)		
44. Verplaatst 30 – 45 meter			63. Loopt gedeelte van trap af		
45. Verplaatst 45 meter of meer; met moeite (struikelt, langzaam voor leeftijd)			64. Loopt gehele trap af; met moeite (langzaam voor leeftijd)		
46. Verplaatst 45 meter of meer zonder moeite			65. Loopt gehele trap af; zonder moeite		
K. Oppervlak buitenshuis					
47. Vlakke ondergrond (asfalt)			**Somscore domein verplaatsen**		
48. Licht oneffend ondergrond (gebarsten stoeptegels etc.)			**Alle items ingevuld?**		
49. Ruw, oneffen ondergrond (gras, grind)			Opmerkingen		
50. Helling op en af					
51. Stoeprand op en af					

Deel 1: Vaardighedenschaal
Sociaal functioneren

A. Begrip betekenis van woorden	*0*	*1*	*D. Complexiteit van expressieve communicatie*	*0*	*1*
1. Oriënteert op geluid			16. Gebruikt gebaren met duidelijke betekenis		
2. Reageert op 'nee'; herkent eigen naam of van bekenden			17. Gebruikt woord met betekenis		
3. Begrijpt 10 woorden			18. Gebruikt 2-3 woordszinnen met betekenis		
4. Begrijpt wanneer u spreekt over relaties tussen mensen of dingen die zichtbaar zijn			19. Gebruikt 4-5 woordszinnen		
5. Begrijpt wanneer u het over tijd heeft en de volgorde van gebeurtenissen			20. Verbindt twee of meer gedachten om eenvoudig verhaal te vertellen		
B. Begrip complexiteit van zinnen	*0*	*1*	*E. Omgaan met problemen*	*0*	*1*
6. Begrijpt korte zinnen over bekende voorwerpen en mensen			21. Probeert probleem aan te geven		
7. Begrijpt enkelvoudige opdrachten met woorden die mensen en dingen beschrijven			22. Vraagt aandacht en moet meteen geholpen worden bij probleem		
8. Begrijpt aanwijzingen die beschrijven waar iets bevindt			23. Zoekt hulp en kan even wachten voordat hulp geboden wordt		
9. Begrijpt samengestelde opdrachten waarbij gebruik wordt gemaakt van als/dan, voor/na, eerste/tweede, etc.			24. Kan probleem en gevoelens beschrijven enigszins in detail		
10. Begrijpt twee zinnen die over hetzelfde gaan maar anders van vorm zijn			25. Bedenkt samen met volwassene oplossing; alledaags probleem		
C. Functioneel gebruik communicatie	*0*	*1*	*F. Sociaal Interactief spel*	*0*	*1*
11. Benoemt personen/dingen			26. Toont belangstelling		
12. Gebruikt specifieke woorden of gebaren om te vragen of ergens op te wijzen			27. Begint uit zichzelf aan bekend spelritueel		
13. Informeert door vragen te stellen			28. Neemt de beurt bij eenvoudig spel wanneer er op gewezen wordt		
14. Beschrijft een voorwerp/actie			29. Poogt de actie van volwassene te imiteren tijdens spel		
15. Verwoordt eigen gevoelens of gedachten			30. Stelt nieuwe stappen voor of reageert op voorstel volwassene		

G. Interacties leeftijdgenoten	*0*	*1*	*J. Tijdsbesef*	*0*	*1*
31. Merkt andere kinderen op, maakt geluidjes of gebaren			47. Algemeen tijdsbesef van maaltijden en dagelijkse handelingen		
32. Interactie tijdens eenvoudige, korte spelmomenten			48. Enig besef van volgorde bekende gebeurtenissen gedurende de week		
33. Probeert eenvoudige ideeën voor spel uit te werken			49. Beschikt over elementaire tijdsbegrippen		
34. Beraamt plannen en speelt samen; enige tijd (tot 30 minuten)			50. Brengt specifieke tijd in verband met activiteit of gebeurtenis		
35. Beraamt plannen en speelt samen; langere tijd (30 tot 60 minuten)			51. Controleert regelmatig de klok of vraagt naar de tijd		
36. Speelt spelletjes met regels			*K. Huishoudelijke taken*	*0*	*1*
H. Spel met voorwerpen	*0*	*1*	52. Begint op aanwijzing te helpen voor eigen spulletjes te zorgen		
37. Manipuleert speelgoed			53. Begint op aanwijzing te helpen met eenvoudige huishoudelijke taken		
38. Gebruikt voorwerpen in "doen alsof" spelletjes			54. Neemt soms initiatief in verzorging van eigen spullen (hulp om taak af te maken)		
39. Voegt materialen bij elkaar om iets te maken			55. Neemt soms initiatief met huishoudelijke taken (hulp om taak af te maken)		
40. Verzint uitgebreide "doen alsof" spelletjes; bekende activiteiten			56. Consequent initiatief en uitvoering van tenminste één huishoudelijke taak; verschillende handelingen en beslissingen		
41. Fantaseert uitgebreide "doen alsof" gebeurtenissen (niet bestaand)					
I. Informatie over zichzelf	*0*	*1*			
42. Noemt eigen voornaam					
43. Noemt eigen voor- en achternaam					
44. Geeft naam en informatie gezinsleden					
45. Kan volledig huisadres opgeven					
46.Kan aanwijzingen geven aan volwassene voor hulp bij thuisbrengen					

L. Zelfbescherming	*0*	*1*	**Somscore domein sociaal functioneren**	
57. Toont gepaste voorzichtigheid bij trappen			**Alle items ingevuld?**	
58. Toont gepaste voorzichtigheid bij Phete of scherpe voorwerpen			Opmerkingen	
59. Hoeft geen aanwijzingen te krijgen bij oversteken straat in bijzijn volwassene				
60. Weet dat het niet mag meegaan of iets mag aannemen van een vreemde				
61. Steekt drukke straat veilig over zonder volwassene				
M. Functioneren in woonomgeving	*0*	*1*		
62. Speelt veilig thuis; geen continu toezicht				
63. In bekende omgeving buitenshuis; periodiek toezicht				
64. Houdt zich aan regels op school of naschoolse opvang				
65. Verkent en functioneert in vertrouwde woonomgeving zonder toezicht				
66. Kan zelfstandig boodschappen doen in buurtwinkel				

Deel 2 en 3: Verzorgerassistentie en hulpmiddelen

Omcirkel de score	Verzorgerassistentie-schaal						Hulpmiddelen-schaal			
	Onafhankelijk	Supervisie	Minimaal	Matig	Maximaal	Totaal	Geen	Kind	Revalidatie	Uitgebreid
ZELFVERZORGING:										
A. Eten: eten en drinken doorsnee maaltijd; sluit uit vlees snijden, potjes, blikjes openen en eten opscheppen.	5	4	3	2	1	0	G	K	R	U
B. Verzorging: tandenpoetsen, haarkammen of borstelen en neusverzorging.	5	4	3	2	1	0	G	K	R	U
C. Wassen: wassen en afdrogen gezicht en handen, een bad of douche nemen; sluit uit transfers in/uit bad of douche, hanteren kraan of het wassen van rug of haren.	5	4	3	2	1	0	G	K	R	U
D. Kleden bovenlichaam: alle kleding voor binnenshuis, sluit uit rugsluitingen; omvat tevens hulp bij aan/af doen van spalk/prothese/corset; sluit uit het pakken van kleding uit kast/la.	5	4	3	2	1	0	G	K	R	U
E. Kleding onderlichaam: alle kleding voor binnenshuis, omvat tevens hulp bij aan/af doen van spalk/prothese; sluit uit het pakken van kleding uit kast/la.	5	4	3	2	1	0	G	K	R	U
F. Toiletgang: kleding, hanteren toilet of gebruik externe hulpmiddelen, en hygiëne; sluit uit toilettransfers, bijhouden wanneer het naar het toilet moet, of verschonen na 'ongelukje'.	5	4	3	2	1	0	G	K	R	U
G. Zindelijkheid urine: controle blaasfunctie dag en nacht, verschonen na 'ongelukje', bijhouden wanneer het moet plassen.	5	4	3	2	1	0	G	K	R	U
H. Zindelijkheid voor ontlasting: darmfunctiecontrole dag en nacht, verschonen na 'ongelukje', bijhouden wanneer het moet poepen.	5	4	3	2	1	0	G	K	R	U
Totalen Zelfverzorging										
	Som score						**Frequenties**			

Omcirkel de score	Verzorgerassistentie-schaal						Hulpmiddelen-schaal			
	Onafhankelijk	Supervisie	Minimaal	Matig	Maximaal	Totaal	Geen	Kind	Revalidatie	Uitgebreid
VERPLAATSEN:										
A. Stoel/toilet transfers: volwassen maat stoel en toilet, rolstoel van het kind.	5	4	3	2	1	0	G	K	R	U
B. Auto transfers: verplaatsen binnen auto, gebruik veiligheidsgordel, transfers en openen en sluiten portieren.	5	4	3	2	1	0	G	K	R	U
C. Bed verplaatsen/transfers: in/uit bed komen en veranderen van houding in het eigen bed.	5	4	3	2	1	0	G	K	R	U
D. Bad transfers: in/uit bad voor volwassenen of douche.	5	4	3	2	1	0	G	K	R	U
E. Verplaatsen binnenshuis: 15 meter (3-4 kamers); sluit uit deuren openen of voorwerpen dragen.	5	4	3	2	1	0	G	K	R	U
F. Verplaatsen buitenshuis: 45 meter op vlak oppervlak; gericht op fysieke mogelijkheid om buitenshuis te verplaatsen (laat buiten beschouwing bereidwilligheid van het kind of veiligheidsoverwegingen zoals bij het oversteken van een straat).	5	4	3	2	1	0	G	K	R	U
G. Trappen: een hele trap op en af gaan (12-15 treden).	5	4	3	2	1	0	G	K	R	U
Totalen Verplaatsen										
	Som score						**Frequenties**			
SOCIAAL FUNCTIONEREN:										
A. Functioneel begrip: begrijpen van aanwijzingen of een verzoek.	5	4	3	2	1	0	G	K	R	U
B. Functionele expressie: mogelijkheid om informatie te geven over activiteiten en behoeften; omvat tevens duidelijk articuleren.	5	4	3	2	1	0	G	K	R	U
C. Gezamenlijk probleem oplossen: omvat communicatie omtrent probleem and samenwerking met verzorger of andere volwassene voor het vinden van een oplossing; bedoeld worden alleen problemen tijdens dagelijkse bezigheden (voorbeeld: zoek geraakt speelgoed, onenigheid over keuze kleding).	5	4	3	2	1	0	G	K	R	U
D. Spel met leeftijdgenoten: mogelijkheid om gezamenlijk activiteiten te plannen en uit te voeren met bekende leeftijdgenoot.	5	4	3	2	1	0	G	K	R	U
E. Veiligheid: voorzichtigheid tijdens dagelijkse onveilige situaties, zoals trappen, scherpe of hete voorwerpen en verkeer.	5	4	3	2	1	0	G	K	R	U
Totalen Sociaal Functioneren										
	Som score						**Frequenties**			

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Curriculum Vitae

Jan Custers werd geboren op 31 maart 1962 te Eindhoven. Hij behaalde in 1980 het HAVO diploma aan het Eckart College te Eindhoven. Van 1980 tot 1984 werd de opleiding Fysiotherapie gevolgd aan de Stichting Utrechtse Paramedische Academie te Utrecht. Van maart 1985 tot januari 1989 was hij werkzaam op de kinderafdeling van het revalidatiecentrum Nederlands Zeehospitium te Den Haag. Van januari 1989 tot oktober 1992 was hij werkzaam op de afdeling fysiotherapie en het team 0-3 jarigen van het revalidatiecentrum de Trappenberg te Huizen. Van oktober 1992 tot heden is hij werkzaam op de afdeling kinderfysiotherapie van het Wilhelmina Kinderziekenhuis / Universitair Medisch Centrum Utrecht (hoofd: prof. dr. P.J.M. Helders).

Specifieke kinderfysiotherapeutische nascholing vond plaats in 1986 met het volgen van de cursus 'Fysiotherapeutische begeleiding van het kind met het MBD-syndroom' (Amsterdam) en in 1987 met het volgen van de cursus 'Neuro Developmental Treatment - Children' (Den Haag). In 1996 werd de 3-jarige Post Academiale Scholing Kinderfysiotherapie aan de Hogeschool van Utrecht afgerond.

In 1991 studeerde hij aan de Open Universiteit en behaalde het certificaat 'Methoden en Technieken van Sociaal-wetenschappelijk onderzoek 1'. Aan dezelfde Universiteit werd in 1993 het certificaat 'Statistiek 1A', en in 1997 het certificaat 'Methoden en Technieken van Sociaal-wetenschappelijk onderzoek 2' behaald.

In 2001 volgde hij een cursus Writing in English for Publication van het Boswell Talen, James Boswell Instituut, Universiteit Utrecht.

De auteur woont in Houten, is getrouwd met Carry Zandstra en heeft twee kinderen: Daan en Lot.

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